Developing psychosocial support for parents and carers of children and young people with an appearance-affecting condition or injury.

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Abstract

Children and young people with appearance-affecting conditions and injuries report common pervasive psychosocial difficulties, regardless of cause or nature of their visible difference. When present alongside the typical challenges of parenting (e.g., practical challenges of childcare, developmental transition points such as puberty and adolescence), parents or carers may also experience psychosocial difficulties related to their child’s visible difference. Current literature is confined to exploring condition-specific concerns of parents, typically in respect to cleft lip and palate (CLP) and burn injuries. Due to the wide range of conditions and injuries which may cause a visible difference, the experiences of parents of many affected children and young people are unknown.

Study 1 took a qualitative approach, utilising individual interviews and focus groups to explore the cross-condition experiences and support needs of parents and carers of children with a variety of visible differences. It was found that parents can experience common psychosocial difficulties regardless of the nature or cause of their child’s visible difference. The qualitative themes highlighted that some parents do experience psychological distress related to their child’s appearance difference. Parents also reported concerns about preparing their child to independently manage challenges related to their condition or injury and lacked clarity about how best to provide support. Study 2 aimed to increase the generalisability of the findings of study 1 and investigate risk and protective factors for parent psychosocial adjustment. The online survey findings aligned with the qualitative data from study 1 and identified several possible cross-condition risk and protective factors.

Despite the cross-condition findings of studies 1 and 2, evidence-informed cross condition support for this population is lacking. Based on the integrated mixed methods findings, the content for an evidence-informed cross-condition intervention was drafted. This content was informed by the Acceptance and Commitment Therapy therapeutic model.

Study 3 took a collaborative Participatory Action Research approach to involve individuals with lived experience of parenting a child with a visible difference and professional advisors. Parents and health and support professionals attended online group workshops and discussed and provided feedback on the content and design of the draft intervention. Finally, study 3 aimed to assess acceptability of the content and design of the intervention. Parents were asked to view the materials and complete an online feedback survey. The findings from this survey indicated that parents found both the content and the presentation of the intervention acceptable.

The studies within this PhD add new knowledge and understanding to the field of visible difference. The intervention output is an innovative and novel evidence-informed intervention for parents of children with a wide range of appearance-affecting conditions and injuries, which aims to promote psychosocial adjustment in this population. A full list of the outputs from this PhD can be found in Appendix A and the intervention materials can be found in Appendix M.

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Contents page

Chapter one: Introduction

This chapter provides an overview of the approach to the PhD and its overall aims. Definitions are provided for important terms used throughout the thesis. To provide context, a brief background on the psychosocial experiences of children and young people with a visible difference is provided. Finally, the structure and outline of the following chapters are described.

1.1. Definition of terms

Visible difference

The term ‘visible difference’ is used extensively throughout this thesis and consequently it is important to clarify how it has been conceptualized. Visible difference has been defined as an appearance which differs from the “norm”. An appearance difference can be caused by a wide range of congenital conditions (e.g., cleft lip and/or palate), acquired injuries (e.g., burn injuries), skin conditions (e.g., eczema), illness or associated treatments (e.g., surgical scarring, chemotherapy). The language previously used to describe visible differences (e.g., disfigurement, abnormality, deformity) has had a negative and stigmatising focus (Rumsey & Harcourt, 2007). Experts in the field and those with lived experience suggest that these terms can be unhelpful and potentially exacerbate the difficulties of those with lived experience (Rumsey & Harcourt, 2004). Although this debate continues to evolve, the term visible difference or appearance-affecting condition and injury is used in this body of work.

These parameters have been previously defined as appearance concerns that do not intersect with a recognised mental health condition (e.g., eating disorders; Rumsey & Harcourt, 2004). This PhD does not include behavioural differences outside of the ‘norm’ within the definition of visible difference (e.g., symptoms of Tourette’s syndrome such as vocal or physical tics). Further details regarding conditions and injuries which may cause a visible difference can be found in Appendix B.

Parent/Parents

Although the 'term' parent will be used throughout this thesis, it is important to acknowledge that a wide-range of individuals take on caring or guardianship responsibilities and may encounter similar experiences to parents. Therefore, for the purposes of this PhD, the term parent will include any adult who has caring or guardianship responsibility for a child with a visible difference.

Child or young person

In the context of this thesis the term "child" or "young person" will be used to refer to any individual who is under the age of 18 and has a visible difference.

The Appearance Collective

The Appearance Collective (AC) is a group of 23 UK-based charitable organisations brought together under a programme of work funded by the Vocational Training Charitable Trust (VTCT) Foundation (www.vtctfoundation.org.uk). All member organisations of the AC support individuals affected by an appearance-affecting condition or injury and their families. The Centre for Appearance Research has developed strong working relationships with the AC charities through research collaborations and the exchange of training and resources.

Participant Pool

The participant pool is a group of individuals who have expressed an interest in taking part in research. When a study is advertised, the individuals in the participant pool are emailed with details of the study and how to take part.

1.2. Background of the PhD

In 2018, the Centre for Appearance Research (CAR) advertised a PhD Studentship with the title, “Providing support and guidance to parent/carers to promote psychosocial adjustment of children and young people affected by appearance-altering conditions and injuries”. This PhD was supported by senior members of AC charitable organisations, who provided letters of support for the original grant application. The clear cross-condition support from these organisations and the ongoing successful collaborations with charities who support individuals with a range of conditions and injuries had an influence on the approach to the research. My experience working with individuals with rare appearance-affecting conditions and injuries also led me to query the provision and support available for conditions of varying prevalence.

In my previous roles of Assistant Psychologist and Research Assistant I worked with children and young people born with genital differences, and their parents. I attended multidisciplinary meetings and clinical appointments with a wide range of professionals including Clinical Psychologists, Clinical Nurse Specialists, Urologists, Gynaecologists, Endocrinologists, and Surgeons. The range of specialist care highlighted the complex physical and psychosocial impact of appearance-affecting conditions. An observation that stood out to me was the lack of attention and support available for the parents who brought their young children in for clinic appointments. Appointments were infrequent (approximately every 6-12 months) and meant that caregivers had minimal support or contact between these short clinic visits, which were focussed primarily on the child’s physical care and treatment. Whilst in this role, I also conducted focus groups with parents to better understand their experiences. It became clear that there was a significant gap in support for their needs, particularly with regards to the psychosocial impact of caring for a child with an appearance difference (Wisniewski, 2017). Consequently, I saw this PhD as an opportunity to further explore the needs of parents of children with a condition or injury which affects their appearance.

My initial interest in the impact of appearance and body image began during my undergraduate degree at Cardiff University, where I had the opportunity to undertake a placement year in a community-based eating disorder service. This placement provided me with time to consider the serious implications of narrow appearance ideals and the detrimental impact they can have on mental and physical health if internalised. After several years working in various mental health services, I began a MSc degree in health psychology at UWE Bristol. This course brought a deeper theoretical understanding to the psychological difficulties I had observed in individuals with long-term health conditions, as well as my own personal challenges of living with a chronic immune disorder. I took this opportunity to bring my new interest in the psychosocial impact of health conditions and appearance together, by conducting a research project on body image with women treated for breast cancer for my MSc thesis. I also became more aware of the work of CAR and the field of visible difference literature. At this point I began clinical work described above and learned more about the experiences and support needs of individuals and families of children with an appearance difference. My applied background in various mental and physical health settings provided me with the skills and experience to focus on identifying unmet support needs and developing appropriate support to address this gap. Therefore, I took a pragmatic approach to the PhD with a focus on creating new knowledge and outputs that would provide support to underserved parents of children with a visible difference.

This PhD provided the opportunity to utilise the experience gained in health psychology research and practice settings and conduct an in-depth investigation into the common experiences of this parent population, influenced by a pragmatic needs assessment approach. In addition, the research process of exploring an evidence-base for future support allowed for the inclusion of individuals with lived experience through public involvement. Therefore, the knowledge and outputs created were informed by the existing literature and theory within the field, the findings from rigorous research conducted in this body of work, and the expertise and knowledge of parents.

1.3. Overall aims

The overall aims of the present PhD were: 1) explore cross condition experiences and support needs of parents of children with a visible difference, 2) investigate possible risk and protective factors for parent psychological distress, and 3) develop support materials to meet the identified cross-condition needs of parents of children with an appearance-affecting condition or injury. To meet these aims two initial studies were conducted with parents and professionals who provided care and support to families. Study 1 and 2 provided insight into the experiences and unmet support needs of this population. Study 2 also provided further understanding regarding the possible risk and protective factors which could be targets for intervention to support parents. Study 3 focussed on the development of a parenting intervention was developed in collaboration with parents and health and support professionals, who provided feedback on the content and design of the draft materials. Finally, a further sample of parents provided initial acceptability data and recommendations for further iterative changes before progression to the next stage of development (Skivington et al., 2021). The next stage of intervention development would be an evaluation of effectiveness, which is beyond the scope of the PhD. The following section of this chapter will provide a brief overview of the structure of the thesis and the content of each chapter.

1.4. Thesis structure and chapter outlines

The following chapter outlines the current literature regarding the experience of caring for a child with a visible difference, the conceptualisation of psychosocial adjustment, parenting and family theory, and the current support provision for parents (Chapter 2). The thesis then critically discusses the theoretical underpinnings and methodology utilised throughout the PhD (Chapter 3). Chapter 4 presents the first study of this PhD which took a qualitative approach, employing one-to-one interviews and focus groups. The cross-condition themes identified in the first study informed the development of a quantitative study. The process of mapping the qualitative data to the quantitative design is described in

Chapter 5. Chapter 6 discusses the online cross-sectional survey study which aimed to generalise the findings of study 1 and investigate possible risk and protective factors for parental psychological distress. The findings from study 1 and 2 were then integrated to draw mixed-methods conclusions. This process of integration is the focus of Chapter 7.

Based on the mixed methods findings, a parenting intervention was developed. Chapter 8 outlines this process of intervention development and discusses the use of a Participatory Action Research approach, the processes and findings of conducting an online acceptability survey, and recommendations for future changes to the intervention. The body of work concludes with a general discussion of the findings in the context of relevant theory, a reflection on aspects of the PhD methodology, and future directions for intervention development and research in the field. The final section of the thesis is comprised of the bibliography of references and appendices.

1.5. Experiences of children and young people

To understand the experiences of parents, it is relevant to acknowledge the experiences of the children and young people themselves. This section considers background literature pertaining to the psychosocial impact of living with a visible difference.

There are no objective measures of visible difference and therefore no precise statistics regarding the prevalence of visible differences. However, it is estimated that approximately one in five people in the UK self-identify as having a visible difference such as a mark, scar, or an appearance-affecting skin or congenital condition, with at least 1.3 million estimated to have a significant visible difference (Changing Faces, 2010). As these prevalence figures are based on self-identification it is important to consider that understanding of the epidemiology of visible differences can be significantly affected by an individual’s own perception of whether their appearance falls within the scope of societal norms.

In an appearance-focused society, an attractive external appearance which adheres to narrow appearance norms is considered ideal and highly prized (Swami, 2012). Westernised appearance norms for women include being thin, tall, having a youthful appearance (Pont et al., 2017; Rossi et al., 2005; Widdows, 2018) and for men include leanness and muscularity (Tiggemann et al., 2008). Furthermore, appearance norms dictate that individuals should have no abnormalities and in body form and a clear and fair complexion (Baumann, 2008).

An altered appearance of any kind that conflicts with current beauty ideals can therefore present significant social and psychological challenges for the affected individual. Furthermore, evidence suggests that living with a visible difference can be particularly challenging for children and young people, as adolescence is a period of social and emotional development characterised by increased value, and investment in appearance and greater social comparison (Stock et al., 2013). Consequently, children and young people with an appearance considered to be socially unacceptable often receive unwanted social attention (e.g., teasing and bullying) and discrimination (e.g., social, or occupational exclusion; Ablett & Thompson, 2016; Feragen & Stock, 2016; Rumsey & Harcourt, 2007).

There is also considerable evidence that many report *common* pervasive psychosocial difficulties and, notably, this is regardless of cause, nature, or extent of their visible difference (Gee et al., 2020; Jenkinson et al., 2015). These include negative self-perceptions, poor body image, low self-esteem, fear of negative evaluation, and anxiety (Wisely & Gaskell, 2012), which can lead to avoidant behaviours (e.g., social withdrawal, Jenkinson et al., 2015). More recently, a survey conducted by the UK charity Changing Faces found that one in three reported feeling depressed, sad, or anxious because of their visible difference (Changing Faces, 2019).

Negative self-perceptions or body image concerns related to a visible difference may also impact a young person's engagement with education. Research with children and adolescents in the general population has indicated that appearance concerns are associated with decline in academic engagement (e.g., attending class, engaging in discussion, paying attention Guimond & Laursen, 2022; Lovegrove & Rumsey, 2004). Changing Faces also conducted a survey which found that 42% of young people with a visible difference felt their appearance affected how they performed at school (e.g., in examinations, Changing Faces, 2017). Respondents also reported that having a visible difference negatively impacted on their experience in the workplace, with 17% reporting that they had left a job due to appearance-related challenges. These findings indicate that living with a visible difference may have a negative impact on social and occupational engagement. However, much of the large-scale empirical evidence related to these outcomes has been conducted with children and young people from the general population. More research is required to better understand the impact of appearance on educational and occupational outcomes for individuals with a visible difference. Nonetheless, overall, there is sufficient evidence to indicate that a visible difference can significantly impact the emotional and social wellbeing of children and young people.

Despite these potential challenges, limited evidence suggests that some individuals adjust and cope well with their appearance-affecting condition or injury. In small sample qualitative studies, some children and young people have reported strategies to manage the challenges of having a visible difference, such as utilising the support of friends and family (Guest et al., 2021; Williamson et al., 2010), whereas others have spoken about coming to accept and appreciate their appearance difference as an important part of their identity (Guest et al., 2021). Therefore, this suggests that adjustment to visible difference can vary between individuals.

1.6. Chapter summary

This initial chapter has introduced the overall aims and approach to this PhD programme of work. Definitions for key terms and language used throughout the thesis have been provided. An overview of the structure and outline of each chapter has been described. Finally, a brief summary of the literature regarding the experiences of children and young people with a visible difference has been discussed. This background sets the scene for the following chapters and research. The next chapter provides a comprehensive review of the current literature pertaining to the experiences of parents, psychosocial adjustment, parenting and family functioning, and the current support provision for parents.

Chapter two: Literature Review

2.1. Introduction

The topic of this PhD intersects with various perspectives within psychological literature (e.g., parenting, health psychology, visible difference), areas that can add valuable contributions to help understand the experiences of the niche group of parents under investigation in this PhD. Therefore, whilst maintaining a focus on the experiences of parenting a child with a visible difference and their related support needs, this chapter also integrates broader and relevant developmental, social and health psychological literature related to the experiences of parenting in general, parenting a child with a health condition and how parents adjust to extraordinary parental challenges. .. More specifically, the literature on psychosocial adjustment from related fields is considered: adjustment to visible difference, adjustment to health conditions, and adjustment to parenting challenges in general. Theoretical perspectives on the nature of parent-child relationships and their relevance to the present PhD are also discussed, before finally examining the current support provision for parents and drawing conclusions about the current limitations of generic parental support.

Conducting a broad review of literature and theory is essential in understanding the existing stance on the experiences of parenthood, and the current provision for support for this population. This enabled the PhD student to build upon the current literature and create a novel contribution to the field. An initial broad literature review was conducted at the beginning of this program of study in October 2018. Further literature reviews have since been conducted to meet the needs of each study. The synthesis presented in this chapter is comprised of these previous literature reviews and an updated search in November-December 2021.

2.2. Parenting a child with a visible difference: the psychosocial impact

As with many parent-child relationships, parents of children with a visible difference provide the majority of care and support for their child throughout their childhood and adolescence. Many appearance-affecting conditions and injuries require parents to take on challenges and responsibilities in addition to the ‘typical’ experiences of parenting (e.g., Nelson et al., 2012; Owens, 2008). Some of these challenges may be comparable to experiences of parents of children with other long-term health conditions. Acute or chronic conditions (appearance-affecting or not) can be viewed as a major life stressor for both the child and caregiver (Compas et al., 2012; Melnyk et al., 2001). Parents of children with a visible difference often encounter many practical and emotional challenges related to managing their child’s condition or injury. Parents can also have difficulties managing their own reactions, whilst attempting to support their child’s adjustment (e.g., (Hawkins et al., 2019; Hlongwa & Rispel, 2018). The following section will explore the role of appearance-related stigma, a key issue for parents to manage, and the practical challenges reported by parents of children with a visible difference.

2.2.1. The impact of appearance-related stigma on parents

Social psychological research has examined the impact of prejudice, stereotyping, and discrimination on members of different social groups (Crocker & Major, 1989; Kite & Whitley, 2016). The literature suggests that individuals may hold negative and stigmatising stereotypes about members of minority groups. This includes racial groups (Brown & Lee, 2005; Hagiwara et al., 2017), individuals with diverse gender identities (Howansky et al., 2019) or sexualities (Carter et al., 2019; Thaker et al., 2019) and individuals experiencing certain health conditions (e.g., Corrigan & Watson, 2002; Earnshaw et al., 2015). Individuals with diverse appearances (i.e. those with noticeable visible differences) that do not meet societal beauty ideals (see section 1.5.) or have a condition or injury that incites disgust or concern in others (e.g., skin condition; Magin et al., 2008) can also be subject to appearance-related social stigma As discussed in section 1.5, many individuals who have visible differences experience social discomfort and perceive social stigma (Jenkinson et al., 2015; Magin et al., 2008; Masnari et al., 2012; Pinquart 2017; Rumsey & Harcourt, 2007; Strauss et al., 2007; Wisley & Gaskell, 2012). In addition, there is the experience of associative stigma among family members; this is perceived stigma arising from their association with the stigmatised individual (Mak & Cheung, 2012; Park & Seo, 2016).

The experience of perceived associative stigma has been reported in family members of individuals with stigmatised conditions (e.g., mental health conditions). A study with 215 parents of a child with a mental health condition found that associative stigma mediated the relationship between symptoms of the condition and care burden for the parents (Park & Seo, 2016). Therefore, associative stigma may act as a mechanism for increased stress for caregivers of stigmatised individuals. Whilst there are relevant challenges shared among those with a range of health conditions and those with an appearance-affecting condition (e.g., decision making, healthcare experiences), it is the experience of appearance-related stigma and appearance-specific concerns that distinguishes those living with or caring for someone with a visible difference from the broader experience of living with a health condition.

Visible difference literature also suggests that parents often experience anxiety related to actual or anticipated social challenges that their child may encounter. For example, parents of children with limb differences have reported experiencing the negative impact of social stigma. An IPA study with seven parents of children with a limb difference found that parents experienced the negative impact of staring and social exclusion from other children (Oliver et al., 2020). Furthermore, a multi-site study carried out in South Africa, found that mothers of children with CLP also described the negative impact of appearance-related stigma (Hlongwa & Rispel, 2018). These mothers described feeling alienated from their communities and unable to take their child out in public for fear of unwanted attention. Parents of children with craniofacial conditions have also reported anticipating difficult social situations, with some parents pursuing "appearance-normalising" treatments to protect themselves and their young child from future social challenges (Feragen et al., 2021).

Consequently, looking different in a society that values a narrow set of appearance ideals can result in pervasive social stigma and discrimination. The phenomena of associative stigma suggests that those close to the individual, such as parents and other caregivers, are also affected, which can lead to social withdrawal or avoidance. Understandably, parents feel anxious about their child navigating the social world and anticipate negative consequences of their child being the recipient of unwanted attention, such as teasing or bullying. This can be emotionally demanding for parents and result in difficult thoughts and feelings related to their child's condition or injury.

2.2.2. Practical and emotional challenges of parenting a child with a visible difference

Many appearance-affecting conditions and injuries can be associated with practical and emotional challenges for parents. For example, the initial diagnostic process or shock following an injury can be difficult, in addition to experiences of stigma. For example, qualitative research with mothers and fathers of children with congenital craniofacial conditions has found that antenatal diagnosis of a CLP can be distressing for parents (Stock et al., 2019; Stock & Rumsey, 2015), and parents of a child with a burn injury have reported psychological difficulties including stress, anxiety and, depression following the injury event (Egberts et al., 2018; Heath et al., 2018a).

Feelings of guilt are also common among parents of children with a visible difference, particularly around the cause of their child's condition. Some mothers report being concerned that behaviour during pregnancy may have caused their child's condition (Chamlin, 2006; Nelson et al., 2009). Whereas fathers have discussed worries about the hereditary nature of the condition (Stock & Rumsey, 2015). Additionally, parents of burn injured children have reported feeling guilt and shame related to the injury events (Hawkins et al., 2019). Notably, these data were primarily collected from White Educated

Industrialised Rich and Democratic (WEIRD) samples (Muthukrishna et al., 2020), thus casual beliefs from minority groups are likely to be poorly represented. For example, there is some limited evidence to suggest that communities in Nigeria attribute the aetiology of CLP to supernatural forces (e.g., evil spirits) or the "will of God" (Olasoji et al., 2007, p.302). Consequently, it is important to consider the limitations of visible difference research related to the experiences of under-represented groups.

2.2.3. Visible difference and additional healthcare needs

Although parents of a child with an appearance-affecting condition have the distinct challenge of managing concerns associated with appearance-related stigma, some of their experiences may be similar to parents of children with other long-term health conditions. Children with appearance-affecting conditions or injuries often have additional healthcare needs that require support from caregivers. This can involve engaging in treatment decision making (Nelson et al., 2012), attending hospital appointments (Feragen et al., 2019; Hlongwa & Rispel, 2018), and carrying out treatment regimens (Lewis-Jones, 2006; Rork et al., 2012). These additional responsibilities can be challenging for parents.

Treatment decision making can be stressful, particularly in the case of elective treatments. For example, qualitative research found that fathers of children with CLP can be especially eager to promote appearance-altering surgery to "normalise" their child's appearance, suggesting an underlying anxiety about their child looking different (Stock & Rumsey, 2015), other mothers and fathers have reported struggling with the decision-making process (Jeffery & Boorman, 2001; Nelson et al., 2012). A study of 35 parents of children with CLP explored the complex process of decision making for elective treatments which aim to "normalise" function, appearance, communication, or identity (Nelson et al., 2012). These authors concluded that social norms about parenting (e.g., feeling a duty to do something), concerns about physical appearance, and healthcare practitioner power had a role in shaping decision making. Once decisions about treatment have been made, parents also felt responsible for and anxious about the outcome of the treatment and whether they were “doing the ‘right’ thing” (Nelson et al., 2012, p.796).

Parents have also reported feeling a challenging mix of emotions following appearance-altering surgery including mourning or sadness, and guilt for exposing their child to post-operative pain in order to change their appearance whereas others have reported overwhelmingly positive emotions (Feragen et al., 2021).. Although this literature provides some insight into the experience of parents of children with craniofacial conditions, there is currently a lack of literature focused on decision-making for invasive appearance-altering treatments for other conditions and injuries (e.g., steroid injections for alopecia). Overall, treatment decision making for parents of children with a visible difference is complex, with many intersecting factors to consider. Regardless of the outcome, this process is emotionally charged and demonstrates an area of challenge for parents caring for a child with an appearance-affecting condition or injury.

Many appearance-affecting conditions and injuries also require long-term management of symptoms or other functional difficulties. In the case of CLP, infants can have difficulties feeding, which requires additional care and can cause distress and anxiety for new parents (Hlongwa & Rispel, 2018; Owens, 2008). Additionally, parents of children with appearance-affecting skin conditions (e.g., eczema) must often carry out extensive time-consuming daily regimes to help manage their child’s symptoms, including bathing and application of topical treatments (Santer et al., 2013). A meta-ethnography of 12 studies on the impact of chronic skin conditions found that parents struggle with other practical aspects when caring for their child such as sleep loss and time off work (Ablett & Thompson, 2016). Parents have also raised practical challenges related to frequent hospital visits for treatment and appointments, which can be costly both financially and in terms of time spent travelling (Heath et al., 2018a; Hlongwa & Rispel, 2018). Parents have reported that these additional pressures or changes to family functioning have been a catalyst for many life changes, including reduced social contact with others (Razera et al., 2017). Razera et al’s cross-sectional study of 100 caregivers of children with CLP found 67% felt they did not have time to care for themselves, due to prioritising their child's care needs. The literature suggests that additional caregiving responsibilities of parents of children can be emotionally and practically demanding and negatively impact quality of life.

2.3. Psychosocial adjustment and visible difference

Although the literature discussed has demonstrated the wide-ranging challenges that can affect parents of children with visible differences, some parents adjust well to their child's condition or injury (Eiserman, 2001; Klein et al., 2006). To understand how this literature can be used to promote psychosocial adjustment, it is first important to clearly define the concept. This section will explore the current literature and theory on adjustment and discuss a definition appropriate for the present PhD.

There is an ongoing debate within the psychological literature regarding what is meant by psychological or psychosocial adjustment. As discussed above, many visible differences are either caused by chronic illnesses (e.g., hair loss caused by alopecia or cancer treatment) or share characteristics with chronic illness (e.g., requiring long-term monitoring and/or treatment). To address this debate in the context of the present PhD, this section begins by exploring psychosocial adjustment to chronic illness more broadly before focusing specifically on adjustment to a visible difference and adjustment in parenting. Psychosocial adjustment in parents of children with a visible difference requires adaptation to complex intersection of challenges (Oliver et al., 2020), and so each aspect of their experience must be considered in-depth.

2.3.1. Psychosocial adjustment to health conditions

It is widely agreed that psychosocial adjustment to long-term health conditions is multifaceted (de Ridder et al., 2008; Stanton et al., 2007). The literature has outlined conceptualisations of adjustment which span multiple domains including cognitive, emotional, physical, behavioural, and interpersonal (Spelten et al., 2002; Stanton et al., 2007). Dependent on theoretical perspective, there is also variation in which components of psychosocial adjustment are prioritised (de Ridder et al., 2008; Taylor; 1983).

Some academics suggest that both positive and negative emotions should be considered (e.g., Stanton et al., 2007), as opposed to a purely deficit perspective or focus on negative psychological experiences which can portray the experience of chronic illness as one defined by “unrelenting suffering” (Stanton et al., 2007, p.568), which is neither a helpful nor accurate representation. It has also been asserted that positive adjustment is not merely the absence of psychological distress, and that positive and negative affect exist on distinct and separate dimensions (Watson et al., 1999) and often co-occur or interact (Folkman & Tedlie Moskowitz, 2000). For example, the broaden-and-build theory of positive emotions has stated that positive affect can work to buffer or repair the impact of negative mood (Fredrickson, 2001). However, it is also important to acknowledge that too much focus on positive experiences may frame adjustment as a goal to attain (Stanton et al., 2007), which could make experiencing difficulties less acceptable. Consequently, a balanced approach that considers both negative and positive affect as indicators of adjustment would be the most appropriate approach, whilst not reinforcing biased stereotypes of responses to chronic illness and associated challenges.

Another important element of psychosocial adjustment to consider is that adjustment is not binary or linear, but a dynamic process. Stage theories of adjustment to major life events (e.g., bereavement) suggest that an individual moves through multiple phases to achieve adjustment (Wortman & Silver, 1989). However, there remains a lack of empirical evidence to support this theoretical perspective. Changing environmental and contextual factors (e.g., support available) mean that individuals may have to readjust to challenging environments (Stanton et al., 2007). The nature of long-term health conditions can result in changes in symptoms or treatments, which will require readjustment (e.g., transient skin conditions such as eczema or psoriasis, Yavuz Daglioglu et al., 2020). Psychosocial adjustment must be considered as an ongoing dynamic process comprised of both positive and negative aspects. This broad understanding of psychosocial adjustment will now be considered in the context of adjustment to visible difference.

2.3.2. Psychosocial adjustment in children with a visible difference

Visible difference experts agree that adjustment is complex and influenced by a wide range of factors (for example see Gee et al., 2020). In response to a lack of understanding of the factors associated with psychosocial adjustment in children and young people with appearance-affecting conditions, Gee and colleagues developed a model to illustrate the predisposing factors and domains that impact psychosocial adjustment in this population (Gee et al., 2020). This model proposed that the following predisposing factors can influence psychosocial adjustment in young people with a visible difference: developmental influences (e.g., developmental life stage), sociocultural influences (e.g., culture/ethnicity), influence of significant others (e.g., level of psychological distress experienced by caregivers), individual characteristics (e.g., optimism, resilience). This model also identified several domains which impact adjustment: 1) psychological wellbeing, 2) social experiences, 3) life engagement, 4) appearance evaluation, 5) treatment/care.

This model considers the influence of key appearance-related cognitions of significant others (e.g., parents) on the adjustment of children and young people with a visible difference (Gee et al., 2020). The level of psychological distress in significant others, level of salience (extent to which self-relevant appearance information is brought to consciousness; Moss et al., 2014) and valence (emotional evaluation of the self in relation to appearance; Moss et al., 2014) of appearance, level of acceptance of their child’s disfigurement, likelihood to endorse gender stereotypes, and level of personal investment in appearance altering surgery were all noted as key predisposing influences. The qualitative themes which informed the development of this model specifically highlight the role of parents who may model maladaptive appearance-focussed attitudes and behaviours to their children. For example, health professionals in Gee et al’s development study stated parents may transfer distress unconsciously onto their child through their approach to treatment and care (e.g., investment in appearance altering treatment). This conceptual model of adjustment suggests that in addition to their own difficulties, parents can influence their child's adjustment through a number of mechanisms. Consequently, it is important to understand the experiences of parents, in order to support both parent and child wellbeing.

2.4. Psychosocial adjustment to the challenges of parenting

Transitioning into the role of a parent or caregiver is a time of significant change for many adults and can be accompanied by a wide range of challenges. As a child grows and develops, parents will continue to encounter challenges related to their caregiving role. Parents of children with a visible difference (whether acquired or present from birth) must manage both these major life transitions and challenges, whilst adjusting to their child's appearance-affecting condition and injury. Furthermore, these challenges can coincide with difficult transition periods in the child's life (e.g., starting school; Feragen et al., 2021), which can complicate further the psychosocial adjustment of parents. To understand the experience of this parent population, it is important to first explore adjustment in the parenting role more broadly.

2.4.1. Individual differences in psychological distress and parenting

Belsky's (1984) theoretical model of determinants of parenting emphasises that even "well-functioning" adults face increased risk of challenges to their wellbeing during this transition. This theoretical position is supported by the literature. Previous systematic reviews have highlighted prevalence rates of between 4.4% and 73.7% of postnatal depression, with the most recent review suggesting a rate of 13% in mothers (Leahy-Warren & McCarthy, 2007). A meta-analysis of 43 studies involving 28,004 parents concluded that the prevalence of post-natal depression was 10.4% in fathers and had a moderate correlation with maternal depression (Paulson & Bazemore, 2010). Therefore, many parents experience psychosocial difficulties when adjusting to their caregiving role.

Further parenting literature has extended Belsky's (1984) model and explored adjustment to parenting as a multifaceted concept related to parental involvement and parent-child relationships (Jia et al., 2016). A study of 182 first-time parents found that individuals suffering from depressed mood struggled with sustained and focussed engagement in enriching activities with their child (Jia et al., 2016). Furthermore, parental anxiety and empathic personal distress ("a self-focussed, aversive reaction induced by observing pain in others" Decety & Lamm, 2013, p.207) was predictive of the amount of time parents spent on childcare. Anxiety and empathic personal distress were significantly positively associated with childcare time in mothers and significantly negatively associated with childcare time in fathers (Jia et al., 2016). These findings suggest that parent psychological adjustment can impact parenting behaviours and there may be gender differences in coping strategies for parenting challenges. Adjustment to the psychological impact of the transition to parenthood is complex and parent behaviours can influence the care received by the child. To further contextualise the interaction between parent adjustment and behaviour and child adjustment and development, a number of theoretical perspectives on parenting will be discussed.

2.5. Interactions in parent and child psychosocial adjustment

Parents and carers are important figures in their child's life. They typically occupy the role of primary caregiver and provide their child with a model of behaviour to observe and imitate. This section explores how parent psychosocial adjustment can influence children within the family unit. This provides important context for the interactions between parent adjustment to their child's visible difference. An awareness of these processes has implications for understanding both parent and child wellbeing.

2.5.1. Parenting styles

The characteristics and implications of individual differences in parenting styles have been widely researched. Baumrind (1966) identified four parenting styles:

authoritative, authoritarian, permissive, and neglectful. Authoritative parenting is characterised by the setting of clear rules and expectations, responsiveness to the child's needs, and open parent-child communication. Authoritarian parents enforced demands through high levels of control and parental power, including the use of threats and punishment. Permissive parents tend to be responsive to their child's needs and are lenient in terms of boundaries and expectations, whereas neglectful parenting is characterised by disengagement from the child and a lack of demandingness and responsiveness to their child's needs.

Characteristics of authoritarian parenting have been identified in research with parents of children with visible differences. Existing literature has found that parents can demonstrate overprotective behaviour and discourage child independence and autonomy (Colletti et al., 2008; Horridge et al., 2010). However, this literature is limited to acquired conditions and injuries which are associated with a traumatic event or diagnosis (e.g., burn injuries and cancer). Nevertheless, individual differences in parenting style could be important in understanding parent and child adjustment to appearance-affecting conditions or injuries.

2.5.2. Family systems theory

Family systems theory (Broderick & Smith, 1979) is a conceptual framework developed by clinicians working in family psychotherapy. It takes a systemic approach, considering the whole family as an interconnected system, which includes smaller sub-systems, including parents, siblings, and parent-child relationships. Family systems theory offers an interpersonal view on family functioning, in which different individuals and sub-systems within the family interact and influence one another (Dallos & Draper, 2015). Each person in the family is seen to influence the others, whose response then influences the first person and so on. These repetitive patterns of influence and interaction are known as circularities (Watzlawick et al., 1967), which encapsulate the feedback loops of family interaction.

This systemic approach provides a more comprehensive understanding of how parenting cognitions and behaviours might influence other members of the family, such as children. This provides some context for why poor psychosocial adjustment in parents of children with a visible difference may also impact negatively on children in the family. However, system models have been criticised for being too reductionist and mechanistic in their explanations (Dallos & Draper, 2015). This model does not account for external sociocultural factors that will inevitably influence family functioning. Therefore, the following section of this chapter will draw on sociocultural models of development to further explore the role of parent-child relationships in psychosocial adjustment.

2.6. Sociocultural models of development

Sociocultural models of development acknowledge that parents, children, and families do not exist in isolation; there are complex internal and external social systems which can influence adjustment. The following models demonstrate the role of socialisation and contextual environments in adjustment. These perspectives are important in understanding the impact of social influence and learning on parent and child adjustment to visible difference.

2.6.1. Bioecological model of human development

The bioecological theory of human development (Bronfenbrenner, 2005) was adapted from the ecological theory of development (Bronfenbrenner, 1992). In this model the family plays an important role in the microsystem context (Bronfenbrenner, 2005). Parents influence the development of the child through their personal characteristics, but also through interactions as part of the proximal processes. Consequently, how parents react, speak about, and manage their child's visible difference will influence their child's adjustment and development. The child also interacts with other microsystems, such as their school environment. The mesosystem illustrates the way in which microsystems interact to affect development. In the context of a child with a visible difference, parent interaction with individuals in other microsystems (e.g., teachers) can be critical for adjustment to their visible difference. Contexts in the exosystem (e.g., media representations, healthcare service) and macrosystem (e.g., cultural beliefs) might also affect the way children adjust to their visible difference. However, this model does possess limitations when explaining the interactions between more distal systems (e.g., the media) and the microsystem. For example, media messages are now present in the family home through the consumption of TV, films, and social media. The bioecological models does not account for these direct interactions of modern family life, which is important to consider as media representation of diverse appearance can be important in shaping child attitudes (Parnell, 2021). Regardless, the bioecological model of development considers many important contextual factors and highlights the essential role of parents within these systems.

2.6.2. Social Learning Theory

Social Learning Theory (SLT) explains the process by which humans acquire behaviours through observation of their environment. Bandura (1977, 1986) proposed that human learning is social in nature and based on the observation and imitation of the behaviour of others. SLT was also later adapted to include more aspects of cognitive processing, including attention paid to certain behaviours (Bandura, 1986). SLT states that associational preferences are important in observational learning. Those who surround the child will determine the behaviour that is repeatedly observed and imitated (Bandura & Walters, 1977). Social learning has been described as “the vehicle by which parents become the catalysts for family-based-behaviours” (Kunkel et al., 2006, p.263). As the parent-child relationship is central to a child’s initial socialisation children and young people can learn a range of behaviours through observation of their parents. Social learning in the parent-child relationship has been observed in numerous behavioural domains, including in relation to paediatric health. For example, adolescent chronic pain-related disability has been significantly positively associated with observable pain behaviours in parents (e.g., restricted mobility; Stone et al., 2017).

A main critique of SLT is that it is reductionist in its account of development and does not acknowledge that human behaviour is often determined by a complex interaction of biological and social factors (Stewart, 2012). However, SLT does provide an understanding of the influence of social learning and provides further rationale for exploring experiences of parents, to support the wellbeing of parent and child.

2.7. Current parent support provision

Building an understanding of the current support available to parents is important to develop an awareness of the context of their challenges and experiences. This section will discuss the various forms of psychosocial support available to parents. As discussed previously (section 2.4.), parents experience fluctuations in adjustment in relation to the "typical" challenges of parenting. It is also common for parents of children with chronic health conditions and visible difference to experience practical and emotional difficulties. This section discusses current support provision for parents of these different groups.

2.7.1. General parent support programmes

Many parenting interventions have aimed to support parents of children with general challenges of parenting. Previous examples of universal parent support initiatives have included the CANparent trial (2012-2015; Lindsay & Totsika, 2017) which offered educational programmes to parents of children 0-6 years old in three geographical areas in England. Targeted programmes of parent support have been designed for children with higher levels of behavioural difficulties than the general population, or at risk of developing such difficulties (Lindsay, 2019). Examples of targeted programmes include Incredible Years (parenting and interpersonal skills training; Webster-Stratton et al., 2004), Triple P Parenting (parent skills training and confidence building; Sanders, 1999), Helping Families Programme (parent training focussed on parent-child relationship, coping strategies, and emotional regulation; Day et al., 2011) and, Empowering parents, Empowering communities (peer-led community training programme, Day et al., 2012). Overall, the evidence indicates that targeted programmes are more effective than universal parenting programmes in improving outcomes such as self-efficacy and mental health (Lindsay, 2019). This may suggest that intervention development is more effective when targeting specific challenges for parents, rather than lower intensity general population initiatives. The existing limited condition-specific literature suggests that parents and carers of children with visible difference may experience specialist appearance-related challenges. Therefore, it may be appropriate to develop a new intervention to meet the needs of this population. To understand the nature of the gap in existing support for parents of children with a visible difference, it was important to review the current provision available for parents of children with a variety of health conditions.

2.7.2. Support for parents of children with chronic health conditions

Support for specific challenges can be observed in parenting interventions targeting parents of children with chronic illness. Interventions for this population can be characterised by variety of content and intensity. Training and support programmes are common for parents of children with long-term health conditions (e.g., Garbutt et al., 2010; Jantzen et al., 2009; Powell et al., 2006). A recent systematic review of skills training interventions for parents of children with chronic health conditions found that these programmes led to increased parent self-efficacy, illness/severity control, child quality of life, and child behaviour (Mitchell et al., 2020), which suggests that training and support programmes may be an effective method of support for parents of children with long-term health conditions.

Psychological support is recognised as important for helping parents to manage the cognitive and emotional challenges of caring for a child with a long-term health condition. A recent systematic review and meta-analysis of psychological interventions (e.g., Cognitive Behavioural Therapy (CBT), family therapy) for parents of children with cancer identified a statistically significant reduction in parental depression following treatment (Bautista et al., 2021). Another review of 44 Randomised Controlled Trials (RCTs) of psychological therapies including 4,697 parents of children with a range of chronic illnesses (e.g., chronic pain, diabetes, asthma) also found that these therapeutic approaches may improve parenting behaviour (e.g., maladaptive coping strategies) and parental mental health posttreatment (Eccleston et al., 2015). However, lack of representation of fathers within trials of parenting interventions remains a consistent limitation (Eccleston et al., 2015), making it difficult to conclude that these interventions are equally effective for both mothers and fathers.

The literature indicates that there are effective specialised parenting interventions for parents of children with long-term health conditions. However, unlike more general parenting interventions, support for these parents must often integrate psychosocial support with ongoing treatment, which can be challenging (Hocking et al., 2014). Parenting challenges, such as child emotional and behavioural development, may intersect with difficulties specifically related to health conditions which increases demand on parent coping resources. Therefore, intervention development and implementation must be conducted with this in mind. These challenges are also present for parents of children with a visible difference, alongside the possible impact of appearance-related stigma. As a result, the support needs of parents of children with a visible difference are likely to be complex. The following section will explore existing support for these individuals.

2.7.3. Existing support for parents of children with a visible difference

Various charitable organisations have developed condition-specific informational resources and advice for parents on various topics including new parent guides, treatment decision-making, and stories from other parents and families (Alopecia UK, 2018; Cleft Lip and Palate Association, 2021; Microtia UK, 2020). Charities who support individuals with a range of visible differences have created informational resources for parents on broader topics such as bullying and teasing (Changing Faces, 2020). Although these resources may be a helpful way for parents to gain information about managing their child’s condition, they are based on anecdotal rather than rigorously obtained evidence and have not been systematically evaluated to assess their effectiveness.

The existing evidence-based support for parents of children with a visible difference is also reasonably limited. A systematic review of 15 studies that evaluated 10 interventions aimed at improving psychosocial outcomes of parents of children with appearance-affecting skin conditions found moderate to strong evidence of the effectiveness of the Triple P Parenting Program and the Early Family Intervention Program (children with CLP) in promoting parent wellbeing (Costa, **Thornton,** et al., 2021). The methodological quality of these studies varied greatly, with six reported as weak. None of the interventions included support specifically on the impact of the appearance aspect for either parent or child, indicating a gap in existing support for this population.

Literature investigating the experiences of parents of children with a visible difference clearly demonstrates that parents can experience significant psychological challenges. Despite this, much of the existing support for this parent population is focussed on skills training to support the management of their child's condition, rather than emotional support for parents themselves. To address the lack of parent-focussed support, a psychoeducational website was rigorously developed by Heath et al.(2019), in collaboration with parents and health professionals, to support parents of burn injured children (www.supportingchildrenwithburns.co.uk). It provides information and advice for parents about how to support their child, whilst also offering stress management guidance for them. Initial acceptability testing of this resource identified that parents and professionals found the content relevant, beneficial, and that they would recommend it to others. This suggests that psychoeducational interventions of this nature may be acceptable to parents of children with a visible difference. However, this existing support remains condition-specific, with limited reach and benefit for the wider visible difference population.

This review of the current support provision for parents provides an important foundation for exploring the experiences and support needs of parents with a range of visible differences. Although valuable resources exist, there are currently gaps in existing care and support, as well as methodological limitations within the evidence-base.

2.7.4. Cross-condition support development

The literature outlined in this chapter demonstrates that research addressing the experiences of and support for parents and children with a visible difference is limited and often condition specific. Nonetheless, evidence from existing condition-specific work suggests there may be significant overlap between parent experiences and challenges across different conditions and injuries; challenges that predominantly centre around having a child who looks ‘different’, whatever the cause. Cross-condition research that includes parents of children with a wide range of visible differences is required to provide evidence for these common experiences and challenges. Findings which support the existence of cross-condition experiences and unmet support needs would provide an evidence base for cross-condition support; an efficient and cost-effective method for delivering psychosocial support to parents of children with a wide range of visible difference, including those resulting from under-researched rare diseases. As it was important to consider that parents may wish to see their exact experiences reflected in condition-specific, rather than generalised, content (a potential barrier to engagement), public involvement feedback on this matter was sought at each stage of intervention development. Thus, while it is acknowledged from the outset that a transdiagnostic approach may limit the potential to identify (and address) any condition-specific support needs of parents, this pragmatic stance aimed to address a gap in appearance-specific support that may even complement condition-specific support materials provided by the third sector.

2.8. Limitations

Although the literature described provides initial insights into the experiences and adjustment processes of parents of children with a visible difference, the limitations within the literature must be acknowledged. Firstly, most of the parenting literature in the visible difference field is condition-specific and largely confined to more prevalent appearance-affecting conditions and injuries (e.g., CLP and burn injuries). Within these condition-specific studies, samples remain selective, often representing subgroups within a condition (scald burn injuries, Heath et al., 2018a). Given the wide variety of appearance-affecting conditions and injuries, this limitation has resulted in a large parent population whose experiences and support needs have not been explored or addressed via research. Due to the lack of research attention given to rarer appearance-affecting conditions, it is also not possible to conduct a comprehensive synthesis of the literature to develop cross-condition conclusions or theory about parent experiences.

A further limitation of the existing literature was the lack of representation of the experiences of fathers and male caregivers. Although some research focuses exclusively on fathers (Stock & Rumsey, 2015), the majority of the condition specific literature explores the experiences of mothers (e.g., Hlongwa & Rispel, 2018). One publication which examined the experience and psychosocial adjustment of parents to their child's CLP found that there were differences in the predictor variables associated with adjustment in mothers and fathers (Stock et al., 2020). This suggests that there may be variability of the experience of psychosocial adjustment of parents and carers of different genders. To fully understand the support needs of parents and carers of children with a visible difference, the present PhD aimed to explore the experiences of both female and male caregivers.

The current support available for parents of children with a visible difference is also limited. Existing interventions focus primarily on practical management of the child's condition (e.g., Morawska et al., 2016), despite clear evidence to suggest parents struggle with their own emotional challenges (e.g., Feragen et al., 2021; Hawkins et al., 2019). Relatedly, there is also a lack of appearance-focussed content within interventions. Considering the potential impact of appearance-related stigma as addressed in section 2.2.1, this seems to be a significant gap in the support for this population. Additionally, information related to managing social stigma and discrimination primarily comes from charitable organisations and is often not theory-informed or evidence-based. As a consequence of the lack of cross-condition research, there is also a significant dearth of interventions for parents of children with rarer conditions. A cross-condition approach to visible difference research and support development could address this gap in the visible difference field.

Within the existing visible difference literature, a variety of measures are used to assess psychosocial outcomes. As this parenting literature is still in its infancy and largely limited to condition-specific experiences, there are currently no standardised measures which capture the specific experience of parenting a child with an appearance difference (e.g., parenting self-efficacy specific to child appearance concerns). This presents a challenge for conducting further research with parents of children with visible differences. Further work is required to identify cross-condition experiences, as well as methods of measurement.

2.9. Conclusion

Parents of children with visible differences can experience significant psychosocial challenges related to caring for their child. These challenges can be related to the impact of actual or anticipated appearance-related social stigma or discrimination. Many visible differences are caused by long-term health conditions or injuries, which are accompanied by additional care and treatment needs. These can also result in difficulties for parents. Psychosocial adjustment to visible difference in children and young people has been explored and theorised, however due to the lack of cross-condition literature and theory, parental adjustment to their child’s visible difference is not well understood. The broader parenting and social development literature provide insights into the bidirectional relationships of influence between parent and child adjustment and coping. This provides further rationale for understanding and supporting the experience and support needs of parents of children with a visible difference. Support for this parent population remains condition-specific and confined to a small number of conditions and injuries, with a focus on informational and practical support. While limited condition-specific research indicates that parents may have similar concerns irrespective of their child's appearance-affecting condition, a cross-condition approach is warranted to further explore and understand the experiences and support needs common to all parents of children with any visible difference.

Chapter three: Methodological considerations

This chapter will discuss the theoretical and methodological background and design of this PhD. It includes an exploration of ontology and epistemology and the use of a mixed methods design. Several elements central to the design and the process of conducting the PhD studies will be discussed. Finally, guidelines for the systematic development of complex interventions will be reviewed and mapped onto the stages of the present PhD.

3.1. Research overview

The main aims of the PhD were to 1) explore the cross-condition experiences of parents of children with appearance-affecting conditions and injuries, 2) investigate risk and protective factors for the experience of distress in parents, and 3) develop support for parents of children with appearance-affecting conditions and injuries. To achieve these aims a mixed methods design was used and four studies were conducted. The first was a qualitative design which utilised two qualitative methods (one-to-one interviews and focus groups). The second study was mainly quantitative, with some open-ended questions included to enhance and clarify understanding of parents' experiences. Study 3 utilised Participatory Action Research (PAR; Baum et al., 2006) to develop an intervention in collaboration with individuals with either a lived experience of caring for a child with a visible difference or of supporting families with a child with a visible difference. The PAR approach will be described in detail in chapter eight. Finally, study 3 employed a primarily quantitative online survey to assess the acceptability of the intervention. The designs and procedures of each of these studies will be detailed in subsequent chapters.

3.2. Ontology and Epistemology

3.2.1. Research paradigms: what is a research paradigm?

Research paradigms are a central concept in social science. Morgan (2007) described a paradigm as "shared beliefs within a community of researchers who share a consensus about which questions are meaningful and which methods are most appropriate for answering those questions" (Morgan, 2007, p. 53). Paradigms are the perspectives that encompass different ways that researchers think and experience the world around them. This includes morals and values which influence the way researchers select and approach research questions, and subsequently design and carry out investigations (Morgan, 2007). Research paradigms have also been described as distinct social worlds, that shape the beliefs of individuals and provide a schema for what is considered to be meaningful and appropriate in research (Morgan, 2007). The following section will outline the characteristics of a spectrum of ontological and epistemological paradigms. Within this discussion, theoretical standpoints and positions that informed or resonated with the PhD approach will be explored in greater depth. The PhD student's own positionality will also be discussed and reflected upon in section 3.5.

3.2.2. Ontological perspectives: realism to relativism

The ontological continuum represents different perspectives on the nature of reality: realism, critical realism and relativism. Realists believe that the world exists independently of the mind (Mackay & Petocz, 2011), with one single reality that is independent of the way humans know about it, known as a 'mind independent truth' (Tebes, 2005). Realism assumes a single knowable truth which we can access through the appropriate application of research techniques (Braun & Clarke, 2013). On the opposite end of the spectrum, relativism assumes that there are multiple realities constructed through social context (Braun & Clarke, 2013). Relativism also maintains that because there is no one single, pre-social reality, what is 'true' differs between contexts. As a result, what we can access and come to know through research only reflects these social constructions of where and how knowledge is generated (Nightingale & Cromby, 1999). Between these ontological positions lies critical realism, which this chapter will address in greater detail.

3.2.3. The epistemological spectrum: Positivism to constructivism

In a world where knowledge is produced from so many different sources, theories of knowledge allow for critique and can highlight what we believe to be truthful and meaningful. Theory of the creation of knowledge, epistemology, posits that a prerequisite of possessing knowledge is that an individual must possess beliefs and that that belief must be true (Pritchard, 2018). The epistemological spectrum encompasses different theoretical standpoints which underlie how researchers, as pursuers of knowledge, understand how truth and reality are constructed.

Positivism (or post-positivism) is at one end of the epistemological spectrum. This worldview is associated with a belief that there is one single reality; a truth that can be discovered through research (Feilzer, 2010). This viewpoint is usually related to quantitative approaches, in which researchers seek an objective knowable truth. Positivism is characterised by a reductionist and deterministic approach to inquiry (Creswell et al., 2011). Alternatively, constructivism, typically associated with qualitative approaches, suggests that there is no such thing as one possible reality (Felizer, 2010). Constructivism states that what we know about reality is a product of the contextual and social systems and discourse in which we reside (Burr, 2003). Knowledge can be viewed as “social artefacts” within their social, cultural, and political environments (Braun & Clarke, 2013, p. 30). A constructivist paradigm lends itself to inquiry that is inductive, wherein individual perspectives inform broader patterns and understandings (Denzin, 2012).

Although these two paradigms dominate methodological and theoretical standpoints in social science (Teddlie & Tashakkori, 2009), the usefulness of the concept of traditional paradigms has been challenged by some authors. Kuhn (1962) argued that paradigms can be interpreted as being prescriptive and lead to exclusion of certain methods. Consequently, Kuhn (1962, p. 24) proposed that paradigms of positivism and constructionism “constrain intellectual curiosity and creativity” and reduce researchers’ ability to access aspects of social phenomena, thereby restricting the ability to explore new phenomena   Within this debate, pragmatism offers an alternative paradigm which instead, focuses on the problem to be researched and the utility of research (Creswell et al., 2007).

3.2.4. Pragmatism as an alternative paradigm: challenging philosophical dichotomy

In contrast to potentially limiting post-positivist and constructionist paradigms, pragmatism accepts that philosophically, there are both singular and multiple realities that can be accessed through research inquiry (Feilzer, 2010). Leading classical pragmatists have posited that all main research paradigms derive from the same approach of seeking truth, whether this be a truth that is an objective representation of a single reality or a relative or subjective truth of multiple realities (Dewey, 1925). Consequently, pragmatism takes an anti-dualist approach and challenges the dichotomy of positivism and constructionism (Rorty, 1999).

Pragmatism distinguishes itself from traditional notions of the purely realist or relativist paradigms of positivism and constructivism. Instead, pragmatism theorises that both objective and subjective inquiries should aim to produce knowledge that best represents reality (Rorty, 1999). Rather than viewing distant paradigms at opposite ends of the ontological and epistemological spectrums, pragmatism conceptualises these dichotomies as continuous and interrelated (Ansell & Boin, 2019). Pragmatism challenges dualism by taking an action and change orientated approach (Ansell & Boin, 2019; Morgan, 2007). Instead of debating how we know and access “the truth”, pragmatism focuses on the pursuit of solving problems with “real world” practical applications (Creswell & Plano Clark, 2007; Dewey, 1925).

Although there are many branches of pragmatism, this PhD is most closely aligned with the work of classical pragmatism. Classical pragmatism as described by one leading philosopher within the movement, John Dewey, will be now outlined and discussed in relation to this PhD.

3.2.5. Dewey’s pragmatism: the concepts of experience and inquiry

Dewey’s conceptualisation of experience centres around two questions: 1) what are the sources of our beliefs? 2) what are the meanings of our actions? Dewey theorised that these questions are linked in a cycle (Dewey, 1925). This theory states that the origin of our beliefs arise from our actions and the outcomes of our actions are then found in our beliefs. Thus, our experiences create meaning by bringing beliefs and actions together.

Dewey believed that experiences must include a process of interpretation. Beliefs must be interpreted to create action and similarly, actions must be interpreted to create beliefs. Much of our experience is interpreted in a relatively automated and effortless manner, which Dewey referred to as habit (Dewey et al., 1998). Habit describes the beliefs we have developed from past experiences, which we can manage with our current resources. Dewey described the process of self-conscious decision making as inquiry, in which thoughtful reflection is required (Dewey, 1998). With regard to all interpretation, Dewey posited that all experience (beliefs and actions) is influenced by our social situations (Dewey, 1998). Therefore, Dewey and other pragmatists have emphasised the importance of the inquiry being rooted in real life, which is inherently contextual, emotional, and social.

In Dewey’s conceptualisation of inquiry there is no distinct boundary between everyday inquiry in life and research. Research is thought to be inquiry-led exploration of difficult situations, which is carried out in a more careful and conscious manner. Dewey’s approach to inquiry involves a five-step systematic system (Morgan, 2014): 1) recognising a situation as problematic, 2) considering the difference it makes to define the problem one way rather than another, 3) developing a possible line of action as a response to the problem,

4) evaluating potential actions in terms of their likely consequences, and 5) taking actions that are felt to be likely to address the problematic situation.

3.3. Pragmatism and mixed methods methodology

As discussed above, pragmatism considers the importance of the utility of research and is focused primarily on problem solving and inciting change (Rorty, 1999). Pragmatists argue that research should not aim to represent 'reality' but provide an account of "how things are" and aim to focus on the usefulness of those findings (Feilzer, 2010).

A fundamental concept in pragmatism is the ability to conduct reflexive research (Morgan, 2007). It is essential that pragmatists ask questions of their research, such as "who and what is it for?", as well as considering the influence of the researcher's own values and beliefs (Feilzer, 2010). With this reflective process at its core, pragmatism first considers the problem under investigation and uses this as a foundation for research design decisions (Teddlie et al., 2010). This is distinct from research based on traditional research paradigms, wherein ontology and epistemology would be queried and interrogated as an initial step in research development. Researchers make active choices based on their beliefs (e.g., what is right or wrong) and feelings. This is likely to be influenced by individual thoughts, as well as social context and background. Dewey described knowledge from inquiry as being the outcome of using a belief in practice. The knower and the knowledge are inseparable and connected by the process of inquiry. Consequently, it is important for researchers to continuously engage in reflection on their own positionality and to remain aware of the influence this may have on the research conducted.

Due to the practical and applied underpinnings of pragmatism, it has been suggested as an appropriate framework for mixed methods research (Feilzer, 2010). This research paradigm supports the use of a mix of research methods to fulfil the aim of conducting applied research and producing socially useful knowledge (Feilzer, 2010). Pragmatism does not require the use of any particular method or combination of methods, nor does it exclude methods (Feilzer, 2010). It aims to examine a particular question or phenomenon and requires the use of the most appropriate research method, or combination of methods, to answer research questions (Feilzer, 2010; Johnson & Onwuegbuzie, 2004). Therefore, when combined with the philosophical assumptions of pragmatism, mixed methods are an optimum approach for applied health research, which allows for flexibility in methods and is guided by the aim to problem solve and produce knowledge with real world impact.

Despite the appropriateness of pragmatism for underpinning the practical, mixed methods approach adopted by this PhD, there are some aspects of the inquiry that are not fully addressed by this alternative paradigm. Pragmatism lacks a critical theoretical viewpoint required to explore experiences of parental and family coping within established, and ultimately, flawed social and health systems (Heeks et al., 2019). Consequently, the perspective of critical realism will also be explored in relation to the current PhD. Critical realism sits on the ontological spectrum between theoretical positions of realism and relativism (Braun & Clarke, 2013). As critical realism is also not positioned at either end of a dualist paradigmatic approach, it has been proposed that critical realism is a good theoretical companion for pragmatism (Heeks et al., 2019). Furthermore, the addition of critical realism strengthens the research by integrating this critical lens. The combination of critical realism and pragmatism with be discussed further in section 3.4.1.

3.4. Critical realism

Critical realism argues the existence of a knowable world accessible through research, which is present behind subjective and socially constructed knowledge (Madill et al., 2000). This position suggests that researchers need to be able to claim that some true reality exists, so that research can elicit beneficial change (Stainton Rogers & Stainton Rogers, 1997).

Roy Bhaskar, a philosopher who initiated the development of critical realism, stated that there is a reality that exists independently from our individual thoughts and this reality can be differentiated into three different levels (Figure 1; Bhaskar, 1989; Houston, 2001).

Firstly, the empirical level consists of experienced events (what we observe happening). Secondly, the actual level which consists of all events, whether we experience them or not (what is actually happening regardless of whether we observe it). Finally, the real/causal level which encompasses the causal mechanisms that generate events occurring at the actual level.

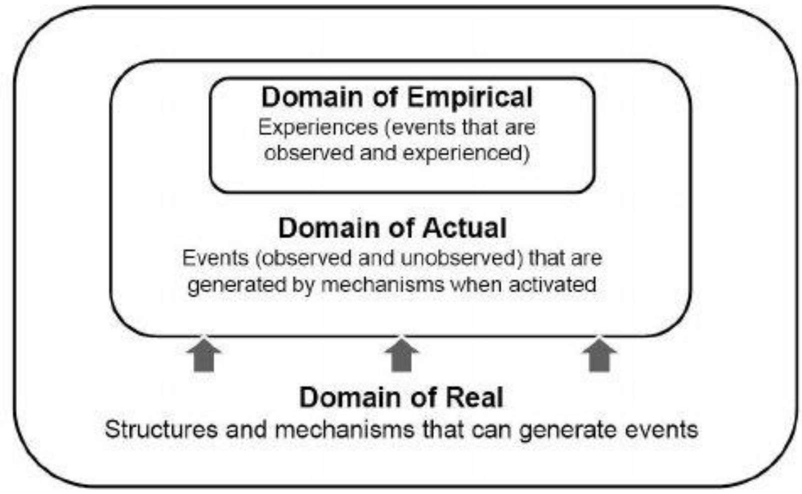

Figure 1: Levels of reality described in Bhaskar's critical realism (Mingers, 2004)

Thus, Bhaskar conceptualised reality as being distinct from and including more than our experiences at an empirical level. As a result, there are elements that we cannot know or access. The real/causal level is central to Bhaskar’s work (Bhaskar, 1989). The causal level of reality is not subject to “direct perception” (Houston, 2001), and so Bhaskar referred to this level of reality as being separate from and “greater than the domain of the empirical” (Bhaskar 1998, p.12). The causal level of Bhaskar’s realism also refers to the existence of concealed mechanisms that contribute to a phenomenon or experience. This core concept of the functioning of the causal level of reality is known as “open systems” (Houston, 2001, p 850). Due to the interactions between these unknown mechanisms, we can only predict probable outcomes or “tendencies” rather than certainties. (Houston, 2001, p.850).

Bhaskar’s critical realism suggests that people’s actions in the social world are influenced by internal and unseen psychological mechanisms, and by wider social mechanisms, which we may not be aware of or able to access (Houston, 2001). This theoretical position acknowledges the interacting roles of social structure (Foucalt, 1972) and individual human agency (Giddens, 1991). Bhaskar proposed that critical realism can be used to think critically regarding existing systems, particularly when these systems lead to oppression. One example related to the present PhD would be the social stigma experienced by children and young people with an appearance outside of societal norms. Houston (2001) suggested that this critical approach should be accomplished through identification of patterns of unmet needs at either an individual or group level. In the case of the present PhD, this mapped onto the identification of unmet needs in parents, to support them in caring for their child.

A critical realist approach allows for an appreciation of the interactions between the physical appearance and functional aspects of visible difference, within the social systems of society, healthcare services, and the family (Williams, 1999). An example of this to consider is that families and couples may behave and interact differently when observed by an outsider, compared to when they are alone. In addition, parents are reporting on their own experience and their interpretation of their child's experience. This process of pre-interpretation is an ontological concept known as hermeneutics (Vandenberghe, 2013). Hermeneutics describe the phenomenon of experience and knowledge being presented to us in a way that has already been interpreted and structured by another person. Therefore, the process of the parents' interpretation is unknown and cannot be accessed by the researcher. A critical awareness of the private systems and processes that occur within the family unit is important to consider when exploring the psychosocial experiences and support needs of parents.

Critical realism is also in line with the use of a mixed methods approach. As previously discussed, Bhaskar describes reality as existing on a number of different levels, some of which we cannot know or access (Bhaskar, 1989; Houston, 2001). This position is consistent with a mixed methods approach that combines qualitative and quantitative methods to represent and explore different facets and layers of the data. A mixed method approach, whilst acknowledging that there will be elements outside perception, can access different voices and viewpoints of reality.

3.4.1. Pragmatist-Critical realism

Pragmatism and critical realism are considered to be appropriate theoretical partners. The intersection of these two positions is referred to in the current literature as pragmatist-critical realism (PCR; Heeks et al., 2019). This combination helps to address this limitation within the pragmatic approach, such as the lack of a critical lens (Simpson, 2018).

Pragmatism and critical realism share a similar objective as alternative options to the traditional dichotomies, in both an epistemological and a methodological sense (Sousa, 2010). PCR combines the practical methodological approach of pragmatism with the established ontological and epistemological underpinning of critical realism (Heeks et al., 2019). As discussed in the previous section, critical realism recognises that reality exists at different levels, some of which we cannot perceive or know. By combining pragmatism with critical realism and acknowledging different levels of reality, PCR provides a more in-depth explanation for the behavioral outcomes of interventions. On the other hand, pragmatism can also enhance critical realism. The applied and action-focused approach of pragmatist methodology complements the theoretical aspirations of critical realism to tackle societal oppression. Therefore, PCR is an appropriate combination of theoretical positions and a suitable approach for the present PhD.

3.5. Reflexivity

The pragmatic approach inquiry described above resonated with my own approach to research. Whilst working in a previous clinical role, I had reflected on the lack of support for parents of children with appearance-affecting conditions. As discussed in section 1.2, I approached this programme of work with a focus on conducting applied and real-world impact. This approach matched closely with the theoretical underpinnings of a pragmatic epistemology. My choice of methodology was also influenced by this pragmatic approach of selecting the most appropriate design to address the problem at hand. I had previously conducted both qualitative (telephone interviews, face-to-face focus groups) and quantitative research (online surveys) but had no experience with mixed methods designs. I could see the independent value of both approaches and understood the strengths that both paradigms could bring to the PhD.

When beginning my PhD, I brought certain beliefs about the experiences of parents and my understanding of the support offered to parents. I also had preconceived ideas about the possible gaps in existing support. Much of the care that I had observed for this parent population followed a medical model of offering surgical intervention, with little ongoing or consistent psychosocial support for the parents or affected child. At this time, I began to consider whether there was psychosocial support available for parents for similar conditions and how psychological and medical care interacted in different paediatric healthcare services.

Having worked in and accessed psychological health services myself for several years, I also came to the PhD project with experience of delivering and accessing a number of different psychological therapies. As a result, I had existing thoughts, feelings, and preferences about different therapeutic models, with the majority of my personal and professional experience being centred around Cognitive Behavioural Therapy. I have had experience of supporting and caring for a parent who experienced significant challenges related to their mental health during my childhood. Therefore, I held beliefs about the potential impact of offering psychosocial support to parents, both on the parent themselves and the children within the family. I also have a close family member with a congenital facial visible difference, who had spoken to me about their own reflections on the way they were parented and experiences they have had, such as interactions with healthcare professionals and comments and attention from the general public. Consequently, I also brought my experience with this family member and my understanding from these conversations to the PhD project.

As outlined in section 3.3., the critical lens of reality adopted by Bhaskar's critical realism was aligned with various aspects of the PhD topic, as well as a mixed methods approach. The critical approach of this position also resonated with my understanding of the way families interact publicly and privately. The interactions and behaviour of a family in their private space reflects the concept of the unknown "open systems" of the causal level of reality, that cannot be known by 'outsiders' to the family group, including researchers and health professionals. Having worked in settings providing child and adolescent mental health support, I held some beliefs of how private and public family systems may differ and the challenges this may pose for exploring the experiences and support needs of parents. My personal and professional experiences had previously led me to think critically about existing systems (e.g., healthcare services). Thus, the critical perspective of this theoretical standpoint aligned with my views about key structures in the experiences of parents.

Finally, it is important to reflect on my own positionality and how my identity may interact with the research process. A researcher's position can be complex and continually shifting as the researcher moves through life. I am not a parent and have no personal experience of raising or caring for a child. I have a minor skin condition with associated scarring. However, this condition developed in adulthood, so I also have no personal experience of being a child with an appearance-affecting condition. In terms of my own family experience, I grew up in a two-parent household until early adolescence, after which my parents separated, and I lived with my mother and younger sister until age 19. As stated previously, it is not possible to entirely separate the researcher from the research. Therefore, transparency about and reflection on my personal positionality is essential in facilitating ongoing reflection and ensuring rigorous and credible research practice.

3.6. Mixed methods research

Mixed methods research has been described as a “workable solution” in the debate between quantitative and qualitative paradigm purists (Johnson & Onwuegbuzie, 2004). Mixed methods research allows researchers to adopt a mixture of quantitative and qualitative methodologies to best serve the aims of their research. Mixed methods research can be distinguished from multimethod research through the tangible integration of data sets in mixed methods, which is not conducted in a multimethod approach (Creamer & Reeping, 2020). Mixed methods research also has no philosophical underpinning in either positivism or constructivism, and instead tends to be rooted in pragmatism (Ortiz & Greene, 2007). Therefore, mixed methods research often has real world applications.

When used alone, quantitative and qualitative methodologies are typically utilised to answer specific types of research question. However, mixed methods research allows for a broader approach and so facilitates the investigation of a much broader range of research questions (Johnson & Onwuegbuzie, 2004). Researchers can be flexible in the questions they can answer when not restrained by methodology of one paradigm. The combination of methods lends itself to the generation and testing of theory, particularly in the case of two stage sequential models (Creswell et al., 2007). Consequently, the mixed method paradigm is ideally positioned to facilitate research outcomes with practical real-world applications.

It is important to acknowledge that mixed methods research does have limitations. Mixed methods research can be challenging for a single researcher to conduct. It requires the researcher to learn both research paradigms and how to mix them effectively (Johnson & Onwuegbuzie, 2004). In addition, methodological purists criticise mixed methods research and maintain that researchers should always work within either a qualitative or a quantitative paradigm (Johnson & Onwuegbuzie, 2004). Quantitative purists believe that inquiry should be objective, whereas qualitative purists believe that socially constructed realities exists and that generalisability to all contexts is not possible or appropriate (Johnson & Onwuegbuzie, 2004). There are some methodological issues in mixed methods research that are still debated by research methodologists (Johnson & Onwuegbuzie, 2004). This includes definitive answers to the question of exactly how to mix paradigms and how to interpret conflicting results from different methods. To ensure rigour in the use of this methodology, the process of integrating findings from mixed methods was therefore reported with transparency using established strategies from the literature (see section 7.3). Mixed methods research also tends to be more expensive and time-consuming to conduct (Johnson & Onwuegbuzie, 2004), a practical issue to consider when designing research projects, especially in situations where time and funding are limited, such as during a PhD studentship. The following section of this chapter will address the rationale for the selection of mixed methods design and how it was employed.

3.6.1. Mixed methods models

There are two common over-arching designs for mixing data: co-ordinated and integrated (Greene et al., 2001). A co-ordinated design requires using multiple methods to collect data sets separately which are mixed at the end of the research, at the stage of drawing conclusions (Greene et al., 2001; Mark et al., 1997). In these circumstances, one data set typically enhances or refines another data set (Greene & Caracelli, 1997). Co-ordinated mixed methods designs fit logically within a pragmatic framework (Greene et al., 2001). Quantitative and qualitative investigations can be conducted separately, in the way that best serves the research question. An advantage of this approach is that findings obtained via different methods can be analysed and then integrated for a broader picture of the phenomenon under investigation (Moran-Ellis et al., 2006). This allows maximisation and respect of the different characteristics and utilities of different methods (Willig & Stainton Rogers, 2008). The sequence of the methods must be selected according to what is most appropriate for the topic and aims of the research. There are two widely used sequential mixed methods models: the explanatory sequential design and the exploratory sequential design (Creswell, 2015; Creswell & Plano Clark, 2007).

In an exploratory sequential design (see Figure 2), qualitative data are collected first in a relatively small sample and then followed up with a quantitative investigation in a larger sample (Creswell, 2015). This is a useful design when the topic under investigation is under-researched because it allows the researcher to explore a phenomenon or problem, before identifying the variables for the quantitative element of the investigation (Creswell, 2015). In this design, the emphasis is placed on the exploratory element of the mixed methods, the qualitative data. As is the intention of the exploratory sequential design (Greene et al., 1989), the quantitative stage of research is developed from and grounded in the views and experiences of participants captured in the initial qualitative data collection. Thus, the quantitative research can then be useful for exploring additional facets of the constructs identified in the qualitative research (Creswell & Clark, 2017). The quantitative research also works as a tool to test for transferability and generalisability of the findings outside of the specific culture of the qualitative sample (Creswell & Clark, 2017).

A sequential exploratory model was selected for the design of the initial two studies. This design was most appropriate due to the scarcity of existing cross-condition literature in this parent population; thus, an exploratory, qualitatively driven approach was required. This allowed for an in-depth exploration of the experiences of parents before the variables for further investigation were identified prior to quantitative investigations (Creswell, 2015; Creswell & Clark, 2017). The quantitative study extended the findings of study 1 by examining additional aspects of the variables and testing for generalisability, whilst remaining firmly grounded in the experiences of participants captured in the qualitative findings.

One challenge of this approach to mixed methods design is that it can be more time consuming than alternative mixed methods approaches (Creswell & Clark, 2017). An additional stage is required between the design and implementation of each piece of research, wherein decisions are made about which findings to expand upon and how to do this. It is also worth noting that two different samples are required for the qualitative and quantitative research. A small purposeful sample was utilised in the first study, followed by a large sample of different participants to enhance the generalisability of the quantitative research. The time taken to conduct qualitative and quantitative research structured in an exploratory sequential design was considered and accounted for in the timeline of the PhD. See Figure 2 for diagram of proposed sequential exploratory model.

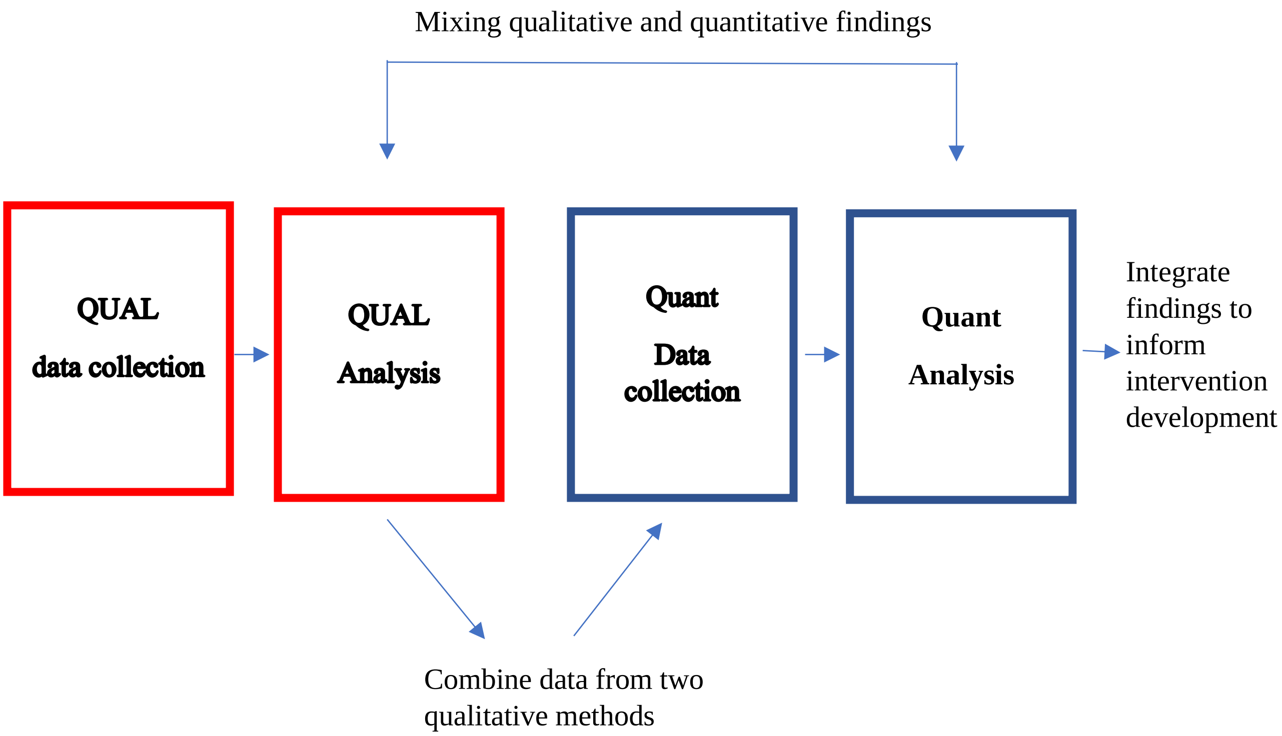

Figure 2: Proposed sequential exploratory design for the first two studies of the PhD

3.7. Assessing quality in mixed methods research

The current literature has proposed that a specialist framework is necessary for the assessment of quality in mixed methods research (Tashakkori et al., 2015). These authors proposed a clear framework to offer guidance to researchers, establish a common language, and provide direction for future development. This section will explore various approaches to quality assessment, the development of a proposed framework and how this framework maps onto the work conducted.

A review of literature concerning the quality of mixed methods research found that the number of publications on the topic have been increasing since 2005, with a particular increase from 2011 (Fàbregues & Molina-Azorín, 2017). In addition, most research addressing mixed methodological issues were from the disciplines of education and health, suggesting that mixed methods research is utilised regularly in these fields.

Bryman et al. (2008) investigated the quality criteria that social policy researchers deemed appropriate for assessing quality in mixed methods research and found that very few respondents believe that traditional quantitative or qualitative criteria should be used (9.2% and 6.3% respectively). Instead, 82.1% of respondents proposed that a combination of traditional and alternative criteria would be more effective (with 2.5% stating "other criteria" should be used). Bryman et al concluded that quantitative or qualitative criteria should not be used in isolation to assess quality in mixed methods research. rather a third criteria combining these two approaches is required, supporting the need for a mixed methods research quality framework.

3.7.1. Acknowledging established criteria

When considering the quality of mixed methods research, many authors will initially refer to the established and accepted criteria for quantitative and qualitative research (Teddlie & Tashakkori, 2009). This can be a helpful reminder of existing criteria before considering how to appropriately assess the quality criteria for a third methodology, mixed methods. O'Cathain (2010) proposed three different approaches for assessing the quality of mixed methods research: 1) generic research approach, 2) the individual components approach, 3) and the mixed methods approach (O'Cathain, 2010).

The generic approach questions whether mixed methods research requires its own quality criteria and instead suggests the application of generic quality criteria for assessment of mixed methods research. However, generic tools have been criticised for being too generalist to be applicable across different research designs (Katrak et al., 2004). The individual components approach posits that the quality of mixed methods can be assessed by judging the quantitative and qualitative elements separately. A limitation of this approach is that it does not appreciate that mixed methods research is more than just the sum of its quantitative and qualitative components (Creswell et al., 2007). Finally, the mixed methods approach aims to assess the quality of mixed methods research as a distinct methodology, rather than breaking down it down into its component parts.

3.7.2. The mixed method approach to quality assessment

Over many years, researchers have made attempts to develop quality criteria for mixed methods research, rather than just focusing on the individual methodological components. Originally, researchers identified 94 quality criteria, 20 of which were specific to mixed methods research (Caracelli & Riggin, 1994). Tashakkori and Teddlie (2003) then proposed a more comprehensive approach expanding on this original model, to include the concept of inference quality, a combination of design quality, and interpretive rigor related to the authenticity of conclusions from the study (Tashakkori & Teddlie, 2003; Teddlie & Tashakkori, 2009). Other researchers built on this approach by suggesting that inference quality should not only be seen as a desired outcome of mixed methods research, but also as a process. Researchers should consider how inferences are drawn, as well as the inferences themselves (Onwuegbuzie & Johnson, 2006).

Other researchers have adopted a “threats minimisation” approach (Creswell et al., 2007, p.145), that focuses on the attention paid to the mixed methods research knowledge base and the transparency of reporting (Creswell et al., 2007; O’Cathain et al., 2008). This requires researchers to consider the potential threats to validity during data collection and analysis. These mixed methods standards require researchers to draw on the mixed methods research knowledge base and be sensitive to the challenges of using their particular mixed methods design (Creswell et al., 2007). Based on these standards, O’Cathain et al. (2008)

developed a set of quality criteria for conducting and reporting qualitative research. These criteria highlight the importance of design, integration, inferences, and transparency.

3.7.3. A quality framework for mixed methods research

The contributions of the researchers mentioned above were utilised to construct a quality framework for mixed methods research (O'Cathain, 2010). The original model devised by Tashakkori and Teddlie (2009) was used as the core of the framework. All the domains of the quality framework for mixed methods research are outlined in Appendix C and includes all the considerations and actions taken to ensure quality in this mixed methods PhD, mapped onto the different domains within the framework. This brings together the theoretical recommendations for conducting robust mixed methods research and the practical steps taken when conducting the studies that comprise this PhD.

3.8. Ethical considerations

All studies received ethical approval from the University of the West of England ethics committee (see study chapters for further details). Discipline specific ethical guidelines were consulted to ensure that the research adhered to good practice within psychological science. The British Psychological Society (BPS) provide a code of conduct for human research ethics, which was referred to throughout the research process. These guidelines are based on four core principles: 1) respect for the autonomy, privacy, and dignity of individuals, groups, and communities, 2) scientific integrity, 3) social responsibility, and 4) maximising benefit and minimising harm (BPS, 2021). The BPS guidelines define risk as "the potential physical or psychological harm, discomfort or stress to human participants that a research project may generate" (BPS, 2021, p.10). Relevant ethical concerns will now be explored, alongside control measures implemented to reduce risk of harm.

The PhD focussed on exploring the experiences of parents of children with an appearance-affecting condition or injury. The topics discussed with parents, including public involvement representatives, during data collection had the potential to be sensitive, challenging, and emotive. For example, parents were often asked to speak broadly about their experience of caring for their child. This required parents to reflect on their initial reactions and experiences to their child's visible difference. Existing research suggests parents' initial reactions to their child's visible difference can include feelings of shock, distress, anxiety, and guilt (Nelson et al., 2012; Heath et al., 2018; Costa et al., 2019; Hawkins et al., 2019). A study of 45 parents of children aged 6 and under with a burn injury found that 28.89% of parents experienced clinically significant symptoms in line with the DSM-IV-TR criteria for post-traumatic stress disorder, including re-experiencing (Odar et al., 2013). Some of the topics raised may have required parents to reflect on or speak about experiences which caused them a high level of distress. Therefore, it is important to consider the possible impact of this research on parents and how to mitigate risks to psychological wellbeing.

All studies were designed with control measures to reduce risk to participants. The research materials were designed to be accessible and provide prospective participants with sufficient information to ensure valid informed consent (BPS, 2021). All participants were also informed of their right to withdraw without consequence, and the limitations of this (e.g., right to withdraw data before up to two weeks following completion of the study). Participants were signposted to possible sources of support (e.g., Samaritans, Mind, Changing Faces) should they need support or information following participation. Any study specific ethical considerations will be reported and reflected upon in the respective study chapters.

3.9. Sampling methods

Given the difficulty in accessing an entire population, research data are typically collected from a subset of the population, otherwise known as a sample (Field, 2009), which is then used to infer about the rest of the population. Consequently, the method of sample selection has important implications for the data collected and the conclusions drawn from the study.

3.9.1. Homogeneity and Heterogeneity in samples

One central concern when considering sampling bias is that a group of participants with certain characteristics will confound the data collected for the study. In some cases, such as this PhD, the researcher is concerned with investigating differences or similarities between categories or groups of people (Coolican, 2018) and so it is important to ensure that these characteristics are represented so that the sample reflects differences in the population under investigation. One way to promote homogeneity and heterogeneity within the sample is to establish clear inclusion and exclusion criteria (Robinson, 2014). The more specific the criteria are, the more homogenous the sample will be.

The inclusion criteria for the samples within the present PhD were parents or carers of children or young people (aged 0-18 years) with a condition or injury that affects their appearance. In terms of homogeneity, all participants were parents or held caring or guardianship responsibilities for a child with an appearance altering condition or injury. However, the samples aimed to be heterogeneous for a number of other characteristics, including type of condition or injury and child age. This was exploratory work which attempted to identify cross-condition experiences of parents of children with different visible differences and of developmental stages. To achieve broad heterogeneous samples, it is important to consider what methods of sampling are most suited to this goal.

3.9.2. Purposive sampling

Purposive sampling is a type of non-random sampling (Brewer & Gregoire, 2009). This method is the deliberate choice of participants due to the qualities the participant possesses (Etikan et al., 2016). Criterion sampling is a purposeful sampling strategy in which participants who meet a certain set of criteria are selected from a larger sample (Palinkas et al., 2015). This method enables researchers to specify a set of characteristics that are determined by areas of interest within the phenomenon. This can facilitate the inclusion of a greater breadth of experiences within the sample. Criterion sampling is deemed suitable for combining aspects of qualitative (depth) and quantitative (breadth) inquiry within mixed methods research sampling (Sandelowski, 2000). To identify the characteristics that would need to be represented in the population targeted within this PhD, a preliminary sampling framework, informed by existing literature and theory from the visible difference and body image field, was developed to guide recruitment (See Figure 3).

3.9.3. Developing a purposive sampling framework

Visible differences are typically conceptualised in terms of whether they are congenital (from birth, Harris, 1997) or acquired (e.g., burn injury, Wisely & Gaskell, 2012). These two categories represent different experiences. Individuals with congenital conditions, and their families, have no memory of their life without the condition (Billaud Feragen, 2012). Whereas acquired conditions and injuries develop or occur at different stages of life. As a result, there may be a "before" and an "after" present in memories of the affected individual and those around them. Another feature that identifies variation in visible differences is whether the condition or injury is permanent or transient. Transient conditions (e.g., psoriasis, alopecia, eczema etc.) in which appearance differences can fluctuate, can lead to unpredictability and uncertainty around progression. This can be distressing and have an impact on patient well-being (Nguyen et al., 2016). Lack of predictability regarding the impact of a condition on appearance may make it difficult to develop coping strategies (Moss, 2005) and therefore, transient conditions should be included alongside permanent appearance differences within this sample. It was also important to consider perceived visibility of the difference from the parents' point of view. Variability in whether the difference is "normally visible" to others (i.e., on hands or face) can lead to variation in the experience of psychosocial adjustment (Moss, 2005). Finally, whether the appearance-affecting condition/injury was associated with a functional limitation or difficulty was considered, as increased focus on body functionality can impact how individuals view their body (Alleva et al., 2015; Avalos & Tylka, 2006). It is possible that body functionality may also affect psychosocial adjustment in parents. Figure 3 shows the sampling framework utilised to guide recruitment and designed to include different facets of caring for a child with a visible difference as discussed above.

It is important to note that the present PhD did not recruit through NHS services. Due to the PhD student's existing professional relationships a number of health and support professionals (e.g., Clinical Psychologists met during Assistant Psychology role), it was not necessary to utilise NHS services to recruit these professionals. In addition, the PhD student invested significant time developing working relationships with members of staff at the Appearance Collective charities (e.g., attending workshops, training, and networking events). This facilitated effective routes to recruiting parents from these communities, without requiring the involvement of NHS services.

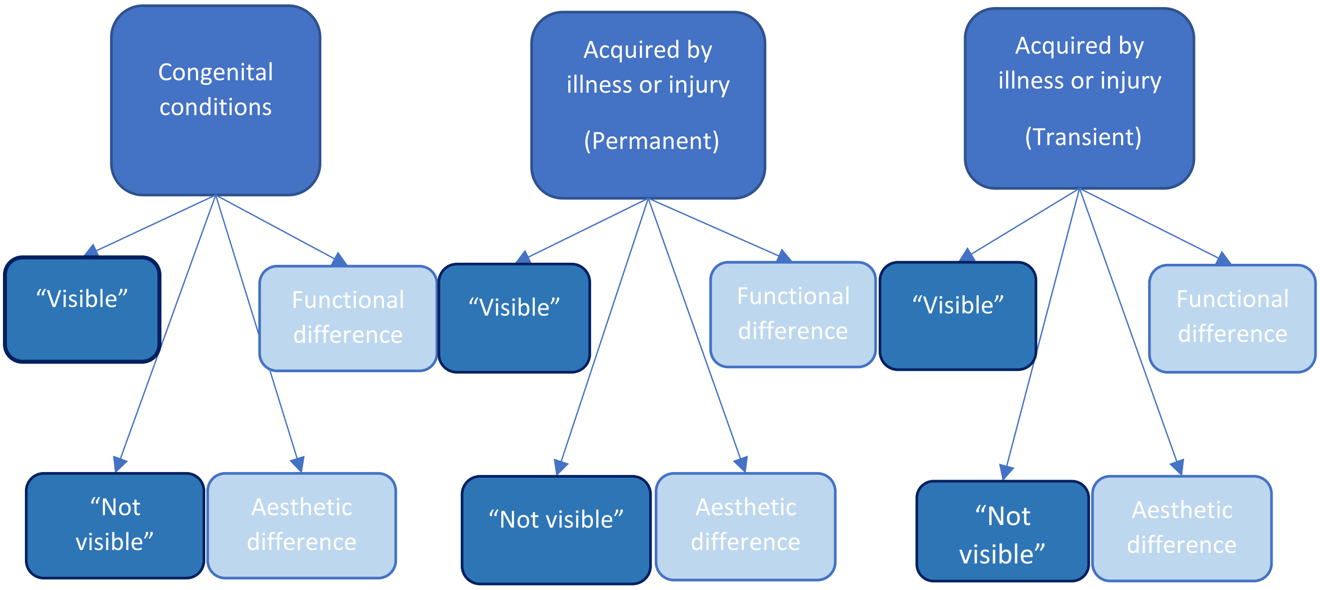

Figure 3: Theoretical sampling framework developed for the present PhD

3.10. Online research design

This PhD employed mostly online recruitment and data collection methods. Although the majority of the recruitment and data collection was designed to be conducted online from the outset, the COVID-19 pandemic eliminated the possibility of any recruitment or data collection being carried out in person from March 2020 onwards. See Appendix E for detailed reflections on the impact of the COVID-19 pandemic. The following section will reflect on the key issues related to online research design.

3.10.1. Online recruitment

Recruitment for studies was conducted online, utilising various online platforms and websites. This was achieved by posting adverts on social media platforms including Instagram, Facebook, and Twitter. These platforms were commonly hosted either by CAR or by member charities of the Appearance Collective. Online forums were also used to reach specific audiences that may be interested in taking part in the research (e.g., *Mumbler* regional parenting forums and *Reddit* condition specific online forums such as, cleft lip and palate, eczema or parenting forums).

Online platforms can be useful for accessing a large number of individuals quickly and with minimal cost (Peer et al., 2017). Online recruitment also has the advantage of reaching a wider demographic of individuals. The resulting samples may be more diverse and representative of the target population when compared to offline methods (Rouse, 2020). However, concerns have been raised about the possible biases that could arise from these recruitment methods (Newman et al., 2021). For example, certain online platforms can be skewed towards certain demographic groups (e.g., lower average age, higher levels of education; Paolacci & Chandler, 2014). There have also been concerns raised about the quality of the data collected. Some authors argue that inattentiveness or non-compliant (Necka et al., 2016) or fraudulent responses (Dennis, Goodson & Pearson, 2020) can pose a risk to data quality. Regardless of these concerns, data collected from online platforms tends to be of good quality; passing attention checks (e.g., length of time to complete research) and scoring highly on measures of reliability (Chandler et al., 2019). For example, a meta-analysis of 90 independent samples, including 32,121 participants, found that data collected from online platforms demonstrated similar psychometric outcomes as data collected from conventional sources (i.e., not using online platforms; Walter et al., 2019).

The use of online recruitment was reflected upon throughout the PhD process. Recruitment strategies were adapted to reduce risk of bias from online recruitment. After reflecting on study 1 recruitment methods (targeting Appearance Collective charities only), a broader range of support organisations and online platforms were approached for studies 2, 3a, and 3b, to reduce any possible sampling bias that may occur from targeting a single organisation or online platform. Intentionally, recruitment for the latter studies also included approaching forums that supported minority groups, to increase the diversity of the sample. To prevent fraudulent behaviour, security measures were added into the survey design (e.g., inclusion of captcha authentication, a security measure to distinguish computers and humans). Data were also screened for inattentiveness (e.g., surveys completed in under 10 minutes were excluded). The possible limitations of online recruitment and how this may impact the findings and conclusions drawn for each study will be addressed in corresponding methods sections.

3.10.2. Online data collection platform

An online survey design was employed for study 2 and study 3 (see chapters 6 and 9 for details about these methods). The survey was hosted on the popular survey building platform, Qualtrics. This software is supported by UWE Bristol, thus data collected on this platform is password protected using university user accounts. Qualtrics has many customisable options for survey design. Previous research has found that the visual presentation of questions within a survey can affect participant responses on domains of speed, accuracy, and response bias, indicating that survey design is an important consideration (Matejka, Glueck, Grossman, & Fitzmaurice, 2016). Qualtrics provides flexibility for presenting survey content (e.g., matrix questions, sliding visual analogue scales, open text entry) and the ability to embed graphics.

Qualtrics allows for complex designs using display and skip logic functions to guide participants through survey items, ensuring participants are only exposed to questions that are relevant to their situation and are not overburdened by irrelevant content, which may reduce participant fatigue. Display logic can also facilitate screening of participant eligibility. If a participant does not meet one or several of the inclusion criteria, display logic can direct them out of the survey with a custom message. This helps to ensure that only eligible individuals complete the survey.

3.10.3. Digital exclusion

The possible impact of digital exclusion is an important consideration when conducting online research Digital exclusion can act as a barrier to both research and healthcare (Greer et al., 2019). In addition, the ongoing COVID-19 pandemic has highlighted and exacerbated the digital divide among certain populations in the UK. Many older adults do not have access to the internet nor engage with online platform (Seifert et al., 2021), and digital exclusion has also been associated with lower levels of education and higher rates of unemployment (Helsper & Reisdorf, 2017). Older parents or carers (e.g., grandparents) and individuals from families with fewer financial resources without access to an internet-enabled device, may therefore be inadvertently excluded from online research. It is important to reflect on possible biases that may result from research conducted exclusively online, and the implications this may have for the generalisability of the findings of this PhD (see section 9.2.1.1.).

3.11. Patient and Public Involvement

The National Institute for Health Research (NIHR) defines Patient and Public Involvement (PPI) as the following:

> "*Research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them. It is an active partnership between patients,*

> *carers, and members of the public, with researchers that influences and shapes research" INVOLVE (2021)*

PPI representatives inform research by providing advice and guidance based on their lived experience. PPI cannot be defined by a single research activity, it takes many forms (e.g., advising on recruitment or research materials) and can operate at many levels within and across the research process (Staley, 2015). PPI can also vary in structure depending on those involved. For example, PPI can be discussions with advisory groups or the involvement of single PPI representatives as co-researchers (Staley, 2015).

The Department of Health states that the inclusion of the active involvement of service users and carers is a key element of high-quality research (Taylor, 2002). PPI representatives can provide insight on appropriateness and sensitivity of research materials, recruitment, and data collection methods (Boote et al., 2002). Another commonly reported impact of PPI is an increase in recruitment rate and breadth of samples (Jagosh et al., 2012; Snape et al., 2014). Recent reviews have also found that the integration of public involvement in research can reduce ethical concerns. PPI representatives can give advice on the relevancy of research topics and improve participants' understanding of research and the experience of participation (Brett et al., 2014; Staley, 2009). PPI representatives have also reported personal benefits from contributing to research. For example, representatives involved in cancer research have reported positive outcomes including increased self-confidence, learning new skills and knowledge, and feeling that they had made a difference (Froggatt et al., 2014).

Following growing conversations around PPI and the "nothing about us, without us" approach (e.g., Paul, 2016), many within research communities view involvement as a right, inherently valuable regardless of impact (Staley, 2015). Consequently, questions around how to best conduct PPI should not focus on "should we do it?" but "what is the best way to do it?" To answer this question, it is-essential to examine processes that lead to impactful PPI. INVOLVE (established by the NIHR to support PPI) has published six UK standards for public involvement (INVOLVE, 2019). These provide a framework for practicing effective public involvement, are adaptable to different research situations and settings and are designed to encourage reflection and learning. These standards provide guidance for how PPI should be carried out but it often remains unclear how this is practically translated into research activities (Staniszewska & Denegri, 2013). See Table 1 for details about how this PhD addressed each standard of public involvement.

Table 1: INVOLE (2019) UK standards for public involvement

Standard		Definition	Present PhD
1)	Inclusive opportunities	Involvement opportunities are accessible (e.g., payment for time and accessible locations) and are reaching the communities who are affected by the research.	Allow for flexibility in involvement to accommodate busy lives of parents. Provide compensation for involvement
2)	Working together	Valuing all contributions and building and maintaining respectful within the group.	Developing relationships within the Parent Advisory Group over a long period of time. Including all members in each stage of research.
3)	Support and learning	Providing public involvement representatives with the skills, knowledge, and confidence they require to engage with research.	Provide public involvement training.
4)	Governance	Public involvement in research governance management and decision making, to increase transparency around the work.	Ask PI representatives to provide feedback on ethical and governance issues within the research.
5)	Communications	Accessible language is essential to communicate with a wider audience about public involvement and research.	All public involvement materials written in non-academic language.

6) Impact	Identify and share the difference made by public involvement, so those involved, and others can understand the benefits and knowledge gained from lived experience insight.	Provide summaries of research and impact of public involvement to those involved.

Staley et al (2015) conducted a literature review to consider how PPI works to shape or create change within our research and concluded that it is researchers themselves who often experience the impact of PPI. As addressed above (Section 3.5.), researchers bring their own thoughts, values, and beliefs to their work. PPI representatives also bring their "knowledge in context" or experiential knowledge. It is the interaction between PPI experiential knowledge and a researcher's positionality that is often challenged through the process of PPI. The result is that researchers then gain a form of experiential knowledge through their interaction and collaboration with PPI representatives. This may be why many researchers refer to PPI as a 'lightbulb moment' or 'reality check' (De Wit et al., 2014). Consequently, the impact of PPI is so variable because each researcher will bring a different positionality. Each PPI representative also brings varied experiences to a range of involvement tasks. It is essential for a researcher to reflect and remain aware of their positionality and the changes that are brought about through PPI.

Although generally accepted as a valuable process, there remain challenges to conducting effective PPI. NIHR commissioned a review entitled the "Breaking Boundaries Review" to investigate the progress of PPI within the organisation (Staniszewska et al., 2018). This review identified practical barriers to conducting PPI in health research. These included a lack of public awareness, limited funding to support relationship building, and issues with reimbursement and payment. An exploratory qualitative study of the experiences of public involvement representatives in cancer research identified several challenges to involvement. These included emotional demands of the work, practical demands (e.g., time commitment), difficulties with inaccessible language, and difficulties identifying the difference their input had made (Froggatt et al., 2014).

There also remains a lack of published evidence of the positive impact of PPI (Tritter, 2009; Staniszewska et al., 2011). Some authors argue that for PPI to gain legitimacy, the impact of PPI in research needs to be formally evaluated (Barham, 2011). Respondents to the NIHR review reported scepticism about the value of PPI and challenges related to inconsistency and lack of clarity in approach (Staniszewska et al., 2018). Additionally, although INVOLVE has provided guidance and standards for practice, these have been criticised for being too generic, and it has been suggested that more training is needed for professionals conducting PPI (Staniszewska & Denegri, 2013). Furthermore, there is still significant progress to be made in order to fully embed PPI into research culture (Staniszewska et al., 2018). Increased evaluation of PPI could provide greater understanding of effective approaches to implementation and underpin training for researchers in the future (Staniszewska and Denegri, 2013). See Appendix D for a breakdown of the impact of PPI on the PhD.

Therefore, although PPI is considered an important and valuable process in research, there remain challenges and barriers to its effective implementation. To address these challenges, the role of PPI will be described transparently, and further reflections will be included in the discussion (see Chapter 9).

3.11.1. PPI in the present PhD

PPI was integrated throughout the PhD, with the goal of enhancing the meaning, relevancy, and appropriateness of research for this population. At the outset, a recruitment call sought members for a Parent Advisory Group (PAG) to act as PPI representatives. Prior to recruitment, each potential PAG member was contacted via telephone for a short discussion about the role of the PAG in the research, expectations regarding contributions by members, and time commitments. Five parents of children with a range of appearance-affecting conditions and injuries and ages were recruited. Variation in conditions and developmental stages was required to represent a breadth of parenting experience. All were mothers and identified as female and White British. A father was recruited but had to withdraw due to family commitments. Two parents had a child with a craniofacial condition, two a child with Alopecia and one a child with facial palsy. All parents had close links to charity organisations that support families of children with a visible difference. The impact of the PPI activities is described in Appendix D.

During the first year of the PhD a PPI workshop was also conducted with professionals from the Appearance Collective charities. The aims of this workshop were 1) to identify any existing support for families of children with a visible difference and 2) to discuss their priorities for the development of future parent support. The priorities identified at this workshop included: confidence building for parents, practical advice on how to manage common challenges (e.g., going to school and managing the reactions of others), emotional support for parents (including how to manage parents' emotions), and signposting to existing support. These priorities were considered throughout the PhD and used to inform subsequent study design (e.g., interview schedule and focus group topic guide, study 1).

Before commencing the research programme, members of the PAG were consulted about the cross-condition design of the research. They also reviewed all study materials, informed and supported recruitment strategies and assisted with the dissemination of the findings for studies 1 and 2. More specifically, several PAG members circulated summaries of the results on their associated charity websites and contributed to research outputs (e.g., an episode for "Appearance Matters! The podcast, https://soundcloud.com/appearance-matters/parenting-a-child-with-a-visible-difference). A member of the PAG contributed directly to the recruitment process by aiding in the creation of a recruitment video for study 2.

Finally, the development of a parenting intervention based on the findings of the first two studies included extensive collaboration with public involvement representatives. This process will be described in depth in chapter 8.

3.12. Development of complex interventions

The existing literature has outlined various approaches to developing health interventions. A systematic review identified eight categories of methods of intervention development. (O'Cathain et al., 2019). This taxonomy of approaches is outlined in Table 2 below. As a mixed methods design was used to develop an evidence-base to inform the development of an intervention for parents of children with a visible difference, in partnership with parent representatives with lived experience, the intervention development approach adopted combined evidence and theory-based and partnership approaches.

Table 2: A taxonomy of intervention development approaches (O'Cathain et al., 2019)

Intervention development approach	Definition
Partnership approach	Intended end users of the intervention participate equally in the decision-making about intervention development.
Target population-centred	The intervention is based on views and actions of people who will use it.
Evidence and theory-based	Intervention based on published research evidence and existing theories.
Implementation-based	Intervention is developed with attention to ensuring it will be used in the real world.
Efficiency-based	Components are tested using experimental designs to select which components which will optimise efficiency.
Stepped or phased	Interventions are developed with an emphasis on following a systematic set of processes.
Intervention-specific	An approach is constructed for a specific type of intervention.
Combination	Existing approaches to intervention development are formally combined.

Complex interventions are characterised as those that contain several interacting components (Craig et al., 2008). When speaking about intervention development, there are several dimensions of complexity. These can include: the range of outcomes, their variability in the population, and the number of elements within the intervention (Craig et al., 2008). According to guidelines developed by the Medical Research Council (MRC) and National Institute Health Research, interventions should be developed systematically and grounded in evidence and theory (Craig et al., 2008; Skivington et al., 2021). A previous iteration of these guidelines were previously applied to inform the development of parenting interventions (e.g., Day et al., 2011) and interventions with visible difference populations (e.g., Williamson et al., 2016). The stages of complex intervention development, as proposed in these guidelines, is outlined in Figure 4.

The original guidelines (Craig et al., 2008, Figure 4) were deemed too reductionist as they only focussed on the "binary questions of intervention efficacy (Skivington et al., 2021). This meant that the guidelines did not consider whether an intervention was implementable, cost-effective, scalable, and transferable across contexts. A new iteration of these guidelines was published in 2021 and consider a broader conceptualisation of complexity (Skivington et al., 2021), and now require researchers to continually reflect and consider core elements at each stage of intervention research (See Figure 5). The application of the updated guidelines to the present PhD are discussed in detail in section 8.1.2.1.

The PhD design was mapped onto the MRC guidelines and focussed primarily on the development stage of these guidelines, with some elements of the feasibility/piloting stage. The first and second studies began by identifying an evidence base and identifying/developing theory. This was achieved by assessing the prediction that parents would experience cross-condition challenges, followed by identification and investigation into risk and protective factors for psychological distress among parents. Study 3 developed a psychosocial intervention based on the findings from studies 1 and 2, in collaboration with PI representatives and health and support professionals with experience of supporting affected families. The intervention was iteratively refined, and the final study evaluated the acceptability of the intervention to parents. The above studies and procedures will be fully described in the following chapters.

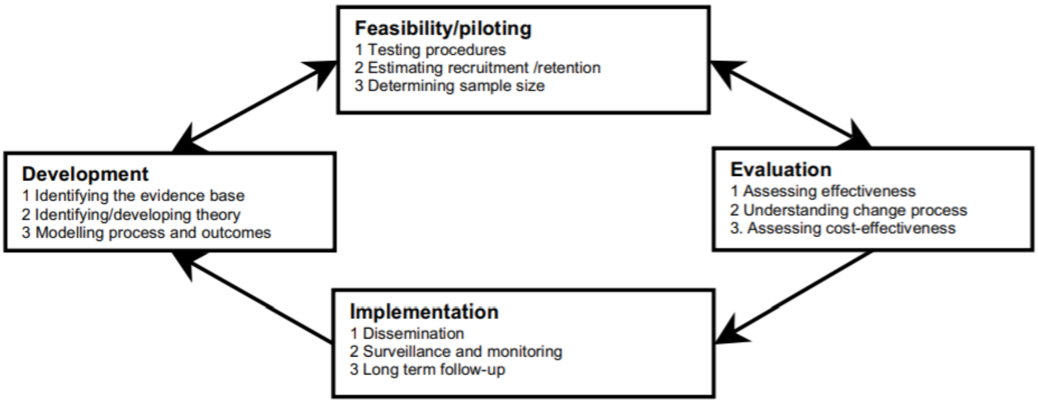

Figure 4: Key elements of the development and evaluation process of complex interventions (Craig et al., 2008)

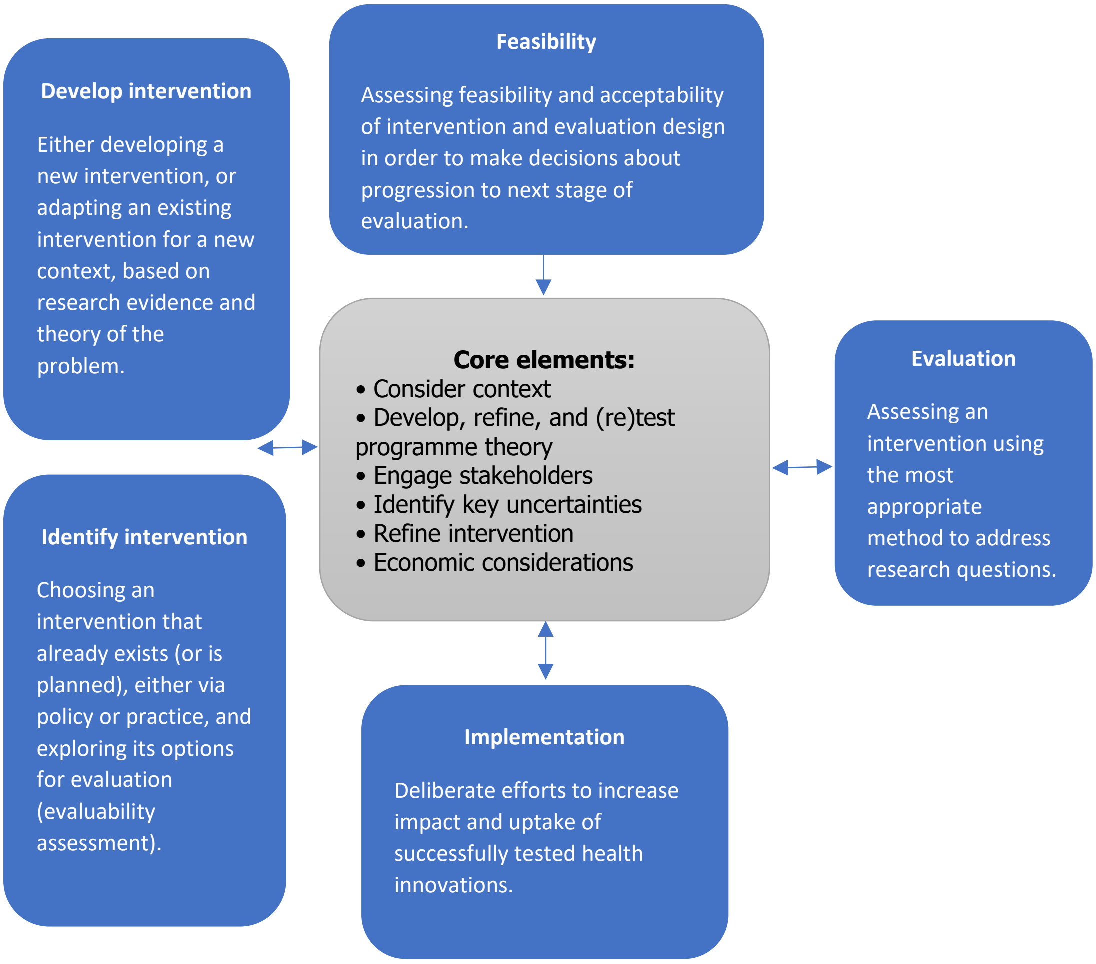

Figure 5: Framework for developing complex interventions (Skivington et al., 2021)

3.13. Chapter summary

This chapter provided a rationale for the methodological approaches selected and an in-depth exploration of pragmatism and critical realism as the theoretical underpinnings for this work. The use of a sequential exploratory mixed methods model was discussed, purposive sampling as the overarching sampling strategy were examined, and the role and importance of PPI in promoting ethical and meaningful research practice was outlined. Finally, this chapter mapped the MRC and NIHR 2019 guidelines for developing complex interventions onto the present PhD studies and processes. The subsequent chapters will now address the specific method and design for each study.

Chapter four: Study 1, A qualitative exploration of the cross-condition experiences and support needs of parents of children with a visible difference

4.1. Introduction

The existing literature exploring the experiences and support needs of parents of children with a visible difference has been confined to condition-specific research with selective samples. Therefore, this study investigated the experiences of parents of children and young people with a range of visible differences and explored whether there are challenges that cut across conditions and injuries, through the perspectives of both parents and health and support professionals. Health and support professionals who work with visible difference have experience with a broad range of conditions and injuries and families from different backgrounds. They also may be less emotionally invested than parents and be able to provide a different perspective on parent experiences. The inclusion of both parents and professionals is also in line with a pragmatic approach to understand the most appropriate methods to rigorously answer the research question.

The study utilised an exploratory qualitative approach to develop an understanding of the experiences of parents and provide insight into whether there is an unmet support need in this population. The following chapter will address the design and conduct of this initial qualitative exploration, which employed interview and focus group data collection methods. Findings will be discussed with reference to literature and theory. The aims and research question for study 1 were as follows.

Research question

What are the cross-condition experiences and support needs of parents of children and young people with a range of appearance-affecting conditions and injuries?

Aims

To explore:

1) The experiences and support needs of parents of children with a visible difference;
2) health and support professionals’ perceptions of the experiences and support needs of parents of children with a visible difference;
3) parent and health and support professionals’ views on the current support provision for parents of children with a visible difference.

4.2. Method

4.2.1. Study design

In line with the proposed sequential exploratory model for study 1 and 2 (see Figure 2), a qualitative research design was selected. Both individual interview and focus group methods were used to collect data. The use of qualitative methods allowed for different perspectives on parent experiences. Individual interviews promoted in-depth conversations about each parent’s personal experience, whereas focus groups provided a format which encouraged interaction and collaboration between parents. Social interactions within focus groups can result in participants asking questions and challenging each other (Braun & Clarke, 2013). Interviews were semi-structured to provide a basis for a systematic exploration of topics using pre-set open-ended questions (DiCicco-Bloom & Crabtree, 2006). This allowed for participant-led exploration, whilst remaining focussed on the key topic.

The aim of employing both interviews and focus groups methods was to capture various facets and voices of parents and the professionals that support them, rather than seeking an accurate truth of their experience. Multiple voices and perspectives introduce another layer of complexity to the data which can then be explored during analysis (Kitzinger, 1995; Parker & Tritter, 2006).

Interview study

Participants were offered different interview formats: face-to-face, skype video call, or telephone. Flexibility with interview format has been shown to increase accessibility of research and improve response-rate (Heath et al., 2018b). All participants opted for telephone interviews, valued for allowing participants to choose a setting where they feel comfortable, which may facilitate disclosure of sensitive information (Novick, 2008), but limited because the researcher has reduced access to non-verbal cues (e.g., body language, facial expressions) and observations of the participants' surroundings (e.g., other people present or distractions; Opdenakker, 2006). Consequently, the PhD student was aware of these constraints and mindful that their understanding of the participant's circumstances may not be complete.

Focus group study

Activity-orientated focus groups were employed. Group activities that are engaging can provide an alternative method for generating discussion between participants and eliciting answers to research questions compared to the traditional question and answer format (Bloor et al., 2001). Activity-orientated questions can be particularly useful to focus the attention of the group on the core topic under investigation (Bloor et al., 2001) and are also appropriate for exploring sensitive topics, which may feel less threatening when addressed through more enjoyable group work (Colucci, 2007).

4.2.2. Research ethics

An ethics application was submitted and received approval in March 2019 with no reviewers' comments to address. For a general discussion of ethical consideration see section 3.8.

4.2.3. Recruitment

Interview study

Member organisations from the Appearance Collective were approached and asked to support the recruitment of parents and professional participants. Charities and organisations were initially contacted by email to inquire whether they would be willing to share information about the research with parents and professionals within their community. An advertisement for the interview study was also posted on the CAR website and shared with the CAR participant pool. Health professionals who had previously shown an interest in research conducted at CAR were also contacted via email. Snowball sampling was also employed to recruit professionals. Participants were asked to share details of the study with colleagues who might be interested.

Focus group study

Caring Matters Now is a charitable organisation that supports individuals affected by Congenital Melanocytic Naevus (CMN, a rare birthmark which can affect appearance of the skin. See Appendix B for further information). This charity contacted CAR to ask the centre to facilitate a parent workshop embedded within their annual conference. It was agreed that a focus group workshop would be run with parents about their experience of caring for a child with an appearance-affecting condition and that data would be collected for this PhD. Caring Matters Now advertised the focus group as part of their conference schedule. This opportunistic data collection session was an effective way of accessing a parent population of children with a rare congenital skin condition. The psychosocial impact of CMN is under researched, especially with regard to qualitative research.

Adequate sample size in qualitative research can be determined using a number of strategies. Data saturation is a concept commonly utilised to determine sample size in qualitative research (Guest et al., 2020). Saturation is observed when no additional new information is identified in additional interviews or focus groups (Sandelowski, 1995). Due to the responsive and interpretative nature of some thematic analysis approaches (e.g., reflexive thematic analysis), there is an ongoing debate about the usefulness of saturation within this method (Braun & Clarke, 2021). However, this study utilised a codebook analysis, rather than a more fluid reflexive style of analysis (Braun & Clarke, 2021). Due to this more structured approach, saturation could be measured and documented through the application of a defined codebook and so could have utility for determining appropriate sample size.

Additionally, the strategy of ‘information power’ in sampling suggests that the more relevant information the sample holds, the lower the number of participants required (Malterud et al., 2016). Based on this concept, sufficient sample size depends on the aims of the study, sample specificity, established theory, quality of dialogue, and analysis strategy. The present study utilised a purposive sampling framework based on criteria informed by the aims of the PhD (see section 3.9.3). This framework informed decision making about adequate information power and sample size within the present study.

4.2.4. Participants

Thirty-one individual interviews were conducted with parents (n=20) and professionals (n=11). The mean age of parent interview participants was 38 years old (SD = 6.1). Most parent interview participants were female and identified as their child’s mother (n=16). Most parents identified as White British (n=16) and married (n=17). The mean age of these parent’s children was 7 years old (SD=4.17) A summary of all demographic information for parent interview participants and their children can be found in Tables 3 and 4. All 11 health and support professionals interviewed identified as female and most also identified as White British (n=7). More demographic information for the health and support professional participants is presented in Table 5. The health and support professional samples included Clinical Psychologists, Charity support staff, and a Dental Surgeon. These professionals provided support to a wide range of conditions and injuries, such as alopecia, ectodermal dysplasia, birthmarks, craniofacial conditions, and burn injuries. More parents than health and support professionals were intentionally recruited and included in the sample. Gaining an in-depth understanding of parents' experiences of caring for children with a wide range of conditions was the primary aim of this study. Whereas professionals work with a wide range of parents and families. Therefore, each individual was able to draw on their experiences with multiple professional cases to provide a breadth of understanding, that also spanned conditions.

Twenty-five parents of children with CMN took part in four focus groups. The mean age of these parents was 46 years old (SD= 6.43) and the majority also identified as female (n=17) and as White British (n=17). The mean age of the children of the parents within the focus group sample was 12 years old (SD=6.01). Further demographic information for this sample is presented in Table 6.

Table 3: Demographics of 20 parent interview participants

	Mean	SD
Age	38	6.1
	N	%
Gender		
Female	16	80
Male	4	20
Relationship to child		
Mother	16	80
Father	4	20
Ethnicity		
Asian other	1	5
White British	16	80
White European	1	5
White Other	1	5
White Scottish	1	5
Marital Status		
Single	2	10
Married	17	85
Divorced	1	5
Highest level of qualification		
GCSEs	2	10
A Levels/HND/BTEC	7	35
Bachelor's degree	7	35
Master's degree	2	10
Doctorate degree/PhD	2	10

Table 4: Demographics of children of interviewed parents

	Mean	SD
Age	7	4.17
	N	%
Child gender		
Female	8	40
Male	12	60
Child condition/ injury		
Alopecia	4	20
Amputee (Meningitis)	3	15
Cleft lip and palate	1	5
Congenital upper limb deficiency	5	25
Eczema	3	15
Facial Palsy	2	10
Microtia	2	10
Vitiligo	1	5

Table 5: Demographic data for 11 health professional interview participants

	Mean	SD
Age	39	8.85
	N	%
Gender		
Female	11	100
Male	0	0
Ethnicity		
White British	7	64
White Scottish	1	9
White Irish	3	27
Highest level of qualification		
A Levels	1	9
Bachelor's Degree	3	27
Master's degree	3	27
Doctorate degree	4	37
Job role		
Clinical Psychologist	4	37
Charity support staff	6	54
Dental Surgeon	1	9

Table 6: Demographics of 25 parents who took part in focus groups

	Mean	SD
Age	46	6.43
Age of child	12	6.01
	N	%
Gender		
Female	18	72
Male	7	28
Ethnicity		
White British	17	68
White Irish	2	8
White European	5	20
Not specified	1	4
Highest level of qualification		
GCSEs	2	8
A Levels/HND/BTEC	5	20
Bachelor's degree	9	36
Master's degree	7	28
Not specified	2	8
Relationship to child		
Mother	17	68
Father	8	32
Gender of child		
Female	17	68
Male	3	12

4.2.5. Materials

Drawing on the existing literature of the psychosocial impact of visible differences, an interview schedule and focus group activities were developed (Hall et al., 2006; Heath et al., 2018a; Jenkinson et al., 2015; Rumsey & Harcourt, 2012; Stock & Rumsey, 2015). The interview schedule and focus group topic guide were developed and reviewed by members of the supervisory team, including a Clinical Psychologist with experience of working with parents of children with a visible difference. This process provided an opportunity to integrate research and clinical perspectives into the development of the materials. Study materials were also reviewed by members of the parent advisory group. See Appendix D for changes made following public involvement feedback. All parents and professionals were provided with detailed information sheets and consent forms prior to taking part in the study.

The focus group activities were developed based on existing visible difference literature, parenting and development theory (e.g., Bronfenbrenner, 1992) alongside preliminary findings from data collected from one-to-one interviews with parents and professionals (completed prior to focus groups). The topics chosen for the focus group activities were based on common themes arising in interview discussions. The focus group discussions were split into four activities relating to the following topic areas: 1) challenges that parents have experienced related to their child’s condition, 2) experiences of social situations, 3) concerns about the future or challenges specific to different life stages and 4) what support they had received as a family or would like to receive as a family. Each activity has a corresponding visual aid produced specifically for this research by the PhD student. These visual aids comprised of key words and line drawings to prompt discussion amongst the focus group participants. Examples of these materials can be found in Appendix F.

4.2.6. Procedure

Interview study

Individuals interested in participating in the research were provided with copies of the participant information sheet and consent form. Once participants had reviewed these documents and agreed that they would be happy to take part, the PhD student arranged a mutually convenient time to carry out the interview. All were conducted via telephone and audio recorded. At the beginning of the interview, participants were asked if they had any questions before beginning the interview and reminded that they could skip a question if they did not want to or felt unable to answer. They were also reminded that they could stop the interview at any point. The interviewer then used the semi-structured interview schedule (see Appendix E) to guide the interview discussion. Mean duration of the parent interviews was 41 minutes, whereas mean duration of the health and support professional interviews was 53 minutes.

Focus group study

The focus group study was conducted during a family conference hosted by Caring Matters Now. Prior to the conference, the focus group was advertised by the charity directors who distributed information sheets. The PhD student led the focus groups and was assisted by two members of the CAR team who, prior to the event, were briefed on the focus group protocol (Appendix E). When parents arrived at the event, they were split into groups based on the age of their child and were asked if they had read the information sheet and to sign a consent form.

The focus group discussions were split into four activities as described above (see section 4.2.5.). Twenty-five parents participated in the focus group discussions. The PhD student introduced each activity in turn. Participants were given an opportunity to ask questions. Participants were provided with flipchart sheets with outlines of the activity to generate discussion points and for them to take notes. Participants led the discussions within their group and each group had a facilitator to help focus the discussion, ask follow-up questions, take notes, and answer any queries. Each activity lasted approximately 10-15 minutes and all group discussions were audio recorded. At the end of the focus group session, the PhD student reiterated the aims of the research to the participants, asked if anyone had questions and left time at the end of the event for parents to approach individually to ask questions. The focus groups were all 60 minutes in duration.

4.2.7. Data analysis

To identify themes that span various conditions a form of thematic analysis was deemed most appropriate. Template analysis is similar to traditional thematic analysis, in that it has a flexible epistemological stance. Grounded theory was considered as an alternative analytic design. However, grounded theory has a less flexible theoretical underpinning and requires sampling for theory construction rather than understanding a broad range of experiences (Charmaz & Thornberg, 2021). Whereas in keeping with a pragmatic approach, template analysis allows for a flexible coding structure that can be adapted to the needs of the study (Brooks et al., 2015). Template analysis uses the structure of a coding template with the option to include a priori themes informed by the potential utility and application of existing ideas, whilst allowing room for the emergence of new codes and themes that answer the question under investigation (Brooks et al., 2015). Additionally, it has been suggested that the discipline of producing the coding template encourages the researcher to take a systematic and well-structured approach to handling the data (Brooks et al., 2015). An interpretative approach was taken to analysing the data. An interpretative approach is concerned with the process of deriving meaning from the data, to gain a deeper insight into psychosocial processes underpinning the behaviour of participants (Willig & Stainton Rogers, 2008). Through this process, the PhD student hoped to develop empathy and understanding of the experiences of participants and uncover meaning beyond the surface level of the data (Willig & Stainton Rogers, 2008).

Template analysis was applied to both interviews (professional and parent separately) and focus groups independently and then integrated through a triangulation protocol (Farmer et al., 2006). Triangulation is a method of combining data sets and is described in greater detail in section 7.3. Integration using a triangulation protocol gives the opportunity to explore agreement, disagreement, and silence between different data sets and perspectives (Farmer et al., 2006). This integration can facilitate a more in-depth understanding of a phenomena by identifying a broader range of voices and concepts (O'Cathain et al., 2010). A limitation of implementing triangulation is that some specific insights from each data set may be explored in less depth within the final themes. However, this analytical strategy is in line with the pragmatic approach to identifying cross-condition experiences and support needs. See Figure 6 for data analysis process. After the data had been coded independently and the triangulation protocol had been applied, a second researcher coded a random selection of transcripts. The coding of the transcripts was discussed, and any discrepancies jointly reviewed and agreed upon.

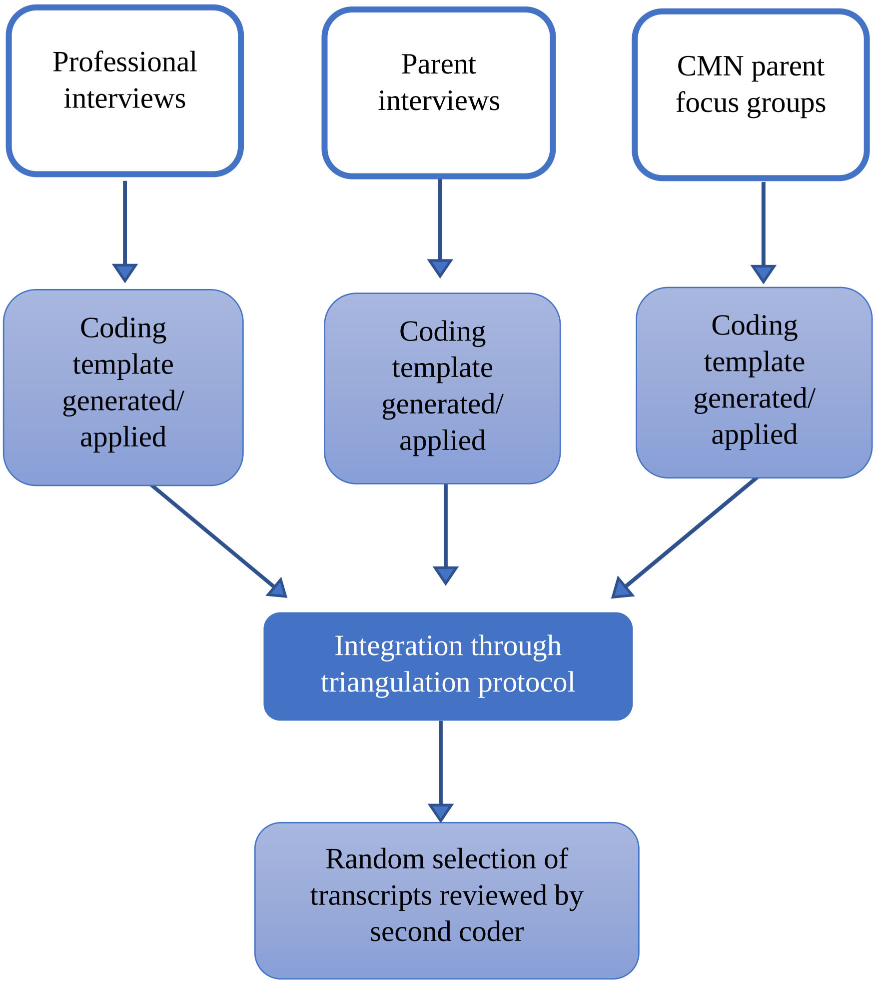

Figure 6. Application of template analysis to mixed qualitative methods design, study one.

4.3. Results

The template analysis identified three main themes. Each main theme also had a number of sub-themes. The main themes were 1) Appearance does(n't) matter, 2) Being 'battle' ready, and 3) Walking the tightrope. These will now be discussed in detail, with exemplar quotes provided. Pseudonyms are used throughout to maintain anonymity. The final coding templates can be seen in Appendix F. A visual representation of the main themes and sub-themes can be seen in Figure 7.

4.3.1. Appearance does(n't) matter

Parents discussed the psychological impact of having a child with a visible difference. As they reflected on this, it was evident that their responses were influenced by the degree to which they themselves valued appearance as an attribute. Many parents were initially distressed by their child's difference, but for some this changed over time after a period of adjustment. For others, the distress persisted. Some parents seemed less concerned about their child's appearance and did not struggle in the same way. Professionals also reflected on parental communication about, and attitudes toward, appearance. When providing support, they found it useful to determine how salient this domain was for the parent.

4.3.1.1. "It broke me"

Parents talked about the distress they experienced related to their child's congenital or acquired visible difference. More specifically, parents discussed initial feelings of shock, anxiety, and low mood in response to seeing the impact of the condition or injury on their child's appearance. For example, Holly described her experience of her 8-year-old daughter losing her hair due to Alopecia:

> *"I kind of have this hope that the first time was the worst time, the real big shell shock, it broke me for about 2 weeks, completely couldn't think about anything else, I was worried about it all the time"*
>
> *Holly, mother of an 8-year-old daughter with alopecia*

Ella spoke about her experience of distress related to her son's condition. She reported hiding her feelings from her son and the guilt she felt for not being able to alleviate his symptoms.

> *"I do to his face but then like when he's gone to bed that's it, I'm like a sobbing mess or you kind of think you know as a mum, you should make it better"*
>
> *Ella, mother of 5-year-old son with eczema*

Many parents equated this experience with feeling "broken". For some, this was a short-term reaction to the initial shock of their child's visible difference. However, for others these feelings persisted for years:

> *"I find it very difficult; I still have some sleepless nights about it because I can just see, it is a horrible thing to deal with"*
>
> *Ellen, mother of 16-year-old daughter with Alopecia*

Parents reflected on how their child's unusual appearance challenged their hopes and dreams of having a "perfect" child, which included them having a conventionally attractive appearance. This often led to feelings of sadness and disappointment and, as Grace (mother of 19-month-old daughter with facial palsy) described, this was often followed by shame and feelings of disloyalty:

> *"I just found it hard that you expect your baby to look a bit of a certain way and obviously she doesn't and it's awful to say, and I feel ashamed to admit it, but I found that quite hard because she didn't visually look as I expected"*

Grace, mother of 19-month-old daughter with facial palsy

Professionals also reflected on parents' feelings of loss related to their child's appearance.

> *"I've heard parents before saying 'this isn't what I wanted for my child', 'I imagine my little girl having beautiful, plaited hair' or whatever so sort of feeling, and then they feeling guilty for feeling like that and actually their child is still the same child, it's just maybe what they thought life was going to be like has twisted on its head"*
>
> *Tara, charity worker, working with families affected by an acquired condition*

Parents also spoke frequently about feelings of guilt associated with the cause of their child's visible difference. For example, Charlotte blamed herself and her body for her daughter's visible difference:

> *"I'd done something wrong, my body had failed my little girl".*
>
> *Charlotte, mother of daughter with CMN in 0-9 year old focus group*

4.3.1.2. "They become so much more"

Despite feeling initially shocked or distressed by their child's visible difference, some parents reported these concerns faded into the background of family life. Others reflected that, ultimately, appearance was not a particularly important aspect of their child's identity. Parents described this as either being able to look past the visible difference, seeing the child as a whole, or not considering the difference to be a significant issue for their child in the first place. Some parents perceived there to be nothing wrong with their child and did not feel the need to alter anything about them. For example, Nina discussed her feelings about her son's condition.

> *"there was no sort of drive inside me to find a cure or anything like that I wasn't, there wasn't something missing that I needed to fix in that way"*
>
> *Nina, mother of 10-year-old son with Alopecia*

Others described how their view of their child's visible difference changed over time. Some parents who had initially experienced distress were able to rebuild and adjust to the new normal:

> *"when the child is born there's quite a lot of sadness or grief about, you know, the fact that their child has a visible difference but over the course of the following years they come to terms with it"*
>
> *Lisa, Clinical Psychologist working with congenital conditions.*

Mollie (mother of 9-year-old son with an upper limb difference) described how her child's ability to cope fuelled this healing process:

> *"Initially when I first had him, and I think it's the same for a lot of the other parents with children with limb differences, you worry about how they will manage and you really quickly learn that they cope amazingly, those kind of worries had kind of disappeared within the first 3 years probably"*
>
> *Mollie, mother of 9-year-old son with an upper limb difference*

As children grew and developed into a young person with likes, dislikes, and a distinct personality, they became defined by multiple attributes rather than just their appearance:

> *"when they're born you feel like they're just a birthmark on a child and then they just become their name and that child and then that funny person and the one that only eats fish fingers and they become so much more, there are just other things that takeover"*
>
> *Rachel, mother of daughter with CMN; 14–18-year-old focus group*

Professionals observed that parents vary in their judgments of the importance of appearance and felt that this affected how parents viewed their child's visible difference. Sarah observed that parents who are more open in their approach to their child's condition or injury seem to adjust well.

> *"parents who cope well are the ones that kind of embrace it in a way and give lots of reassurance to their child that it doesn't matter what they look like"*
>
> *Sarah, charity worker, working with acquired conditions*

Some parents also described broader positive outcomes associated with their experience of having a child with a visible difference. These included appreciating that they now had greater acceptance of appearance diversity, empathy for those who are different and awareness of their own, and their family's' resilience.

> *"it's certainly made my husband and I more, maybe accepting and trying to be more understanding of differences and I think it helps us to teach both of our kids to look beyond outer experiences and accept that people have differences and differences are good".*
>
> *Caitlin (mother of 4-year-old son with Microtia; a congenital condition which affects the development of the external ear)*

4.3.2. Being "battle" ready

Parents vocalised their concerns about their child coping with their difference, drawing on various social situations they have experienced and framing these as potential threats or barriers that their child would need to overcome. Parents were cognisant of the need for children to manage social challenges associated with their visible difference independently (e.g., addressing comments made about their appearance). Parents and professionals discussed how parents approach 'arming' their children for these challenges. However, after shielding their child and being responsible for managing the impact of the visible difference, parents reflected on having to 'let go' and allow their maturing child to manage on their own. Regardless, parents felt a strong sense of responsibility to ensure that their child is prepared and "battle" ready. Parents felt this concern about the future regardless of whether they perceived that their child was currently coping well or not.

4.3.2.1. Identify the threats

When discussing the challenges that their child might face, many parents worried most about the social impact of the child's visible difference. Parents were concerned about children socialising within their own circles (e.g., at school or with friends), as well as interacting with the public.

Parents were concerned about how others will react to their child and whether an unusual appearance will result in social stigma, for example unwanted negative attention or discrimination, and the negative impact on their child's well-being. They worried that their child might experience social stigma or discrimination and were fearful about the negative impact that might have on their child's wellbeing. Jade was worried her son would be a target:

> *"kids will be kids and kids don't really need an excuse to have a target of other children for bullying and things like that, I'm worried that he's going to become a target"*
>
> *Jade, mother of 6-year-old son with Vitiligo*

Similarly, Mollie summarised a common occurrence reported by many parents of children with a visible difference:

> *"you get a lot of double takes shall we say, a lot of staring, people tend to fall into two categories of staring and not saying anything or just being really forward and saying, 'what happened to him?'"*
>
> *Mollie, mother of 9-year-old with an upper limb difference*

Professionals also shared examples of parents' concerns about the potential negative social impact of their child's appearance. Professionals and parents also described how caregivers often anticipate future, sometimes unknown, threats (e.g., teasing at school). Professionals highlighted that these fears can drive parents to seek appearance-altering treatments.

> *"they're concerned that when these children go to pre-school and when they start in big school that they're going to get teased or bullied or whatever and so they're keen sometimes to try and get more normal looking dentition in advance of that"*
>
> *Ruth, Paediatric dentist working with congenital conditions*

Isobel (mother of son with alopecia) had similar concerns and reflected most parents' desire for their child to fit in and be accepted*: "my overriding thing was that he's going to get picked on at school and that was my concern and you know you want your kids to be happy, you want them to excel in whatever they can and you want them to do well but actually I just wanted, I just want them to fit in and you want them to have an easy life"*

Professionals and parents also described how parents often anticipate future, sometimes unknown, threats. Alice illustrated this by discussing the frequency to which parents "fast-forward" through their child's life to identify these potential threats.

> *"they think oh gosh this is going to be the worst thing ever for my child, and it's fast forwarding, you could have a child who is I don't know 7 years old and has lost their hair and that parent is fast forwarding to high school, their fast forwarding to dating, their fast forwarding to the child never getting married, it's amazing how often that kind of crops up as a scenario"*
>
> *Alice, charity worker working with acquired conditions*

Several parents also discussed gender differences, asserting that appearance is more salient for girls, and reaffirming societal expectations that appearance is closely tied to worth and value for girls/women and less so for boys/men. As a result, parents of girls appeared more concerned about the potential social impact of their child's visible difference.

> *"I'm just worried it may damage her self-esteem and I suppose in so far as being female as well, because I just know that perhaps unfortunately there is a greater emphasis on physical appearances in that respect and maybe she'll be ridiculed more"*
>
> *Sam, father of 10-month-old daughter with an upper limb difference*

4.3.2.2. Shielding my child

Some parents considered attempts to shield their child from potential threats by altering their child's appearance to fit in within appearance norms, for example, by concealing or hiding the visible difference of others:

> *"I am a bit more conscious of it and trying to make sure he keeps covered up but then there are times when we are away here just in a swimming pool, he's quite exposed and it does seem to be when he's in a swimming pool that the spots seem more prominent, I don't know if it's the chlorine, just makes it quite obvious"*
>
> *Jade, mother of 6-year-old son with vitiligo*

> *"he's got really long blonde hair and part of the reason I have it long is because of his microtia"*
>
> *Marie, mother of 2-year-old son with microtia*

Others stepped into social interactions, to offer protection. In some situations, the parents who took actions to conceal their child's visible difference were the ones who expressed negative perceptions of their child's appearance. For example, Ellen stated her concerns about her daughter's "unattractive" appearance:

> *"I can't lie to her and say she looks attractive without a wig because she doesn't, areata I think is particularly unattractive because it's patchy"*
>
> *Ellen, mother of 16-year-old daughter with alopecia*

Of these parents, some reflected that perhaps they were shielding their children too much, but still found it difficult to let go of protective behaviours. Martin articulated this challenge:

> *"I find as well sometimes you can step in too quickly, at the swimming pool you have kids coming up to her saying, 'what are the spots on your skin?' And I'd be stepping in and saying something and actually it's better to let her say something so I had to learn to step away"*
>
> *Martin, father of daughter with CMN, 14–18-year old focus group*

Professionals reported some in-depth reflections on parental motivation to conceal differences. They suggested that parents might choose concealment as a strategy to manage their own vulnerability about their child's visible difference.

> *"I can see that the mother has used a wig as a way of almost to protect herself basically from her own feelings around the child's hair loss basically, in not having to confront that themselves, I've heard a parent in the past tell me "I can't bear to see her without her hair"*
>
> *Alice, charity worker working with an acquired condition*

4.3.2.3. Arming my child

Parents talked about the need to arm their child with psychosocial tools to self-manage challenges. They discussed the process of equipping their child with these tools, either as a strategy in addition to concealing the visible difference, or as a lone strategy. Parents discussed building general confidence and resilience in their children to protect them from difficult social situations and described coping skills they wanted to nurture or instil:

> *"it's trying to arm him with enough mental fortitude around it to be able to deal with those questions and understand that people will have questions and understand"*
>
> *Robert, father of 6-year-old son with limb difference*

There was also an underlying uncertainty felt by parents who queried whether they had done enough:

> *"have we done enough confidence wise to get them through?"*
>
> *Stuart, father of son with CMN, 10-13-year-old son with CMN*

Some parents emphasised the need to develop coping skills, rather than focussing on medical treatment to alter their child's appearance. In these cases, parents prioritised arming their child with strategies to deal with potential threats rather than attempting to shield them.

> *"even now that we're always working on her confidence and her resilience and stuff like that so that she can deal with it rather than going down the route of medical treatment,"*
>
> *Holly, mother of 8-year-old daughter with alopecia*

Many parents also raised the issue of communication with their child about their visible difference and human diversity more generally. They expressed a sense of responsibility for ensuring these conversations informed their child's understanding of their own difference:

> *"we really tried to make him aware of the fact that he has got a little arm, differences are good, everybody's different, daddy is a diabetic and injects himself, mummy's got blonde hair, and you can see differences in everybody, everybody is different, this is one of things that make him different"*
>
> *Robert, father of 6-year-old son with limb difference*

Parents also expressed a sense of responsibility for passing on this information and helping to develop a positive narrative around the visible difference for their child:

> *"one of those questions you dread as a parent of a child like him and I just remember my heart sinking and thinking right I've got to deal with this properly and*

> *not lie really and so I said no your arm unfortunately won't ever grow, it will always be a little arm, but that's ok because your little arm is special"*
>
> *Mollie, mother of 9-year-old son with a limb difference*

Professionals also reflected on discussions with parents who feel a responsibility to pass on condition-specific knowledge that will help their children construct a narrative around the cause and nature of their difference. This they believed was particularly important for children with congenital conditions who do not themselves possess the memories or information around the early years of their condition.

> *"It's important that, especially for example in cleft, those early surgeries happen, that the children themselves don't really hold that history and they are allowed to build that narrative, so the parents pass over that knowledge"*
>
> *Amy, clinical psychologist*

Parents also talked about the importance of modelling adaptive behavioural responses to their child's visible difference. Isobel decided not to explore treatments for her son's alopecia because she did not want him to feel that it was something that needed "curing". She felt that had she pursued treatment there would be a dissonance between her behaviour and the message she was trying to convey to him: that appearance is not the most important thing.

> *"if I'm turning around saying it doesn't matter it's only hair, yet I'm taking him somewhere where I'm trying to cure it, it doesn't really fit in with the of what I'm telling him"*
>
> *Isobel, mother of 13-year-old son with alopecia*

Professionals concurred and suggested that parents who exemplify the use of adaptive approach to manage challenging situations provided children with behaviour to observe and imitate as they began to interact and socialise independently.

> *"if the parent was able to say "oh it's a birthmark" or whatever the condition was "she's not in pain" and then sort of move the conversation along then often the child then learnt oh well that's how my mum fields these questions and I can deal with that too and they seem to be the ones that are more confident and were less concerned about their appearance"*
>
> *Bella, clinical psychologist working with congenital conditions*

> *"it's a similar thing with the wigs like if they're losing their hair then they maybe don't really know how they should feel about it and they look to their parents and if the parents are like oh god we should cover up then the child is kind of like god right this is something terrible, I need to cover up myself"*
>
> *Sarah, charity worker working with acquired conditions*

4.3.3. Walking the tightrope

Parents were concerned about how to protect their children from perceived threats and how much attention to focus on their child's visible difference. Parents considered whether they should try to minimize the impact of the difference and worried that talking about it too much might create a problem where one did not exist. On the other hand, if they avoided the issue then children would lack an observable behaviour to model, preventing them from developing their own narratives. Participants described walking a fine line in attempts to "get it right" whilst also being unsure about where that line lay. Getting it wrong was regarded as potentially disastrous because their child's mental health was at stake.

4.3.3.1. "Messing about in the dark"

Many parents felt lost regarding the best way to support their child and reported that little guidance was available.

> *"It's like everybody is just messing about in the dark, hoping that these things are going to work"*

Holly, mother of 8-year-old daughter with alopecia

Conversations about appearance were particularly difficult for parents, who lacked confidence in their ability to effectively support their child.

> *"it's difficult to use the right words that don't come across as making a judgement about, the way he looks or the way he, his abilities are"*
>
> *Caitlin, mother of 4-year-old son with Microtia*

Striking the balance between too much and too little appearance talk was also challenging:

> *I don't know whether I do dismiss it a bit too much, you know do underplay it a bit too much"*
>
> *Erica, mother of 12-year-old son with limb difference*

This lack of clarity and confidence was also observed by professionals in their conversations with parents. Tara spoke about parents worrying about whether the support they provided was the best for their child. Parent's lack of confidence was reflected in Tara's conversations with parents who expressed anxiety that they might be doing more harm than good.

> *"I think that's the kind of things that parents worry about like is what I'm saying doing more harm or more goods? They're not really sure, should they be encouraging them to cover it or should they be encouraging them to embrace it, those are the questions that we hear from parents"*
>
> *Tara, charity worker working with acquired conditions*

4.3.3.2. The double-edged sword of support

Parents described mixed experiences of reaching out to those they perceived as experts (e.g., professionals or others with experience of the condition). Although some experiences were positive, others were often disappointing.

> *"to this day I remember what she said because we went and we got diagnosed through the GP with a dermatologist, actually at hospital and as when left I sort of said "what can I do?" and they said "there's nothing you can do, it won't kill her"*
>
> *Ellen, mother of 16-year-old daughter with alopecia*

Professionals also talked about a lack of confidence when working with those affected by appearance-affecting conditions; they too were unsure about how to address appearance concerns:

> *"They're nervous that they don't want to get too involved"*
>
> *Ruth, paediatric dental surgeon.*

Parents also sought information and support independently via charity websites and social media, but again with mixed results. Parents found online peer support platforms useful for sharing experiences with other similar parents, which helped to reduce their feelings of isolation.

> *"it's nice to see that other people on these Facebook pages, there's no negativity, there's no like "well you shouldn't be doing thing, you shouldn't be doing that, everyone seems to be really positive about it all, like sharing opinions"*
>
> *Marie, mother of 2-year-old son with microtia*

However, both parents and professionals recognised that peer support could also fuel anxiety exposing parents to worst case scenarios and stories about previously unknown threats and future challenges, further confirming their need to equip their child with coping strategies but with no more knowledge of how to do so.

> "*I sort of read a lot of the conversations in there, some of that is really scary because you get all the worst case scenarios where people go on to lose all their hair so in my head I was preparing for her to lose all her hair, which obviously didn't happen"*
>
> *Holly, mother of 8-year-old daughter with alopecia*

> *"when they've got a new born baby with a condition and when they look at images of what a 10 year old with those syndromes look like or 15 year olds, or adults, I think they're probably really met with the challenges that lie ahead when they see that those children"*
>
> *Lisa, Clinical Psychologist working with congenital conditions*

4.3.3.3. "On the edge of a big black hole"

Parents felt fully responsible for their child's ability to cope with challenging situations and carried the guilt if their child struggled to adjust. The primary concern was that living with a visible difference would affect their child's wellbeing and mental health.

> *"I do obviously worry about his emotional state and he mainly teenage years are my biggest worry and he has over the last 4 years, had problems with anxiety and kind of angry melt downs that kind of thing"*
>
> *Mollie, mother of 9-year-old son with limb difference*

Health professionals also witnessed examples of parents expressing guilt when their child struggled to manage common challenges resulting from looking different. The weight of this responsibility and the severity of the perceived consequences for their child sometimes has a detrimental effect on the psychological wellbeing of the parent. Paige spoke about a mother who was struggling to cope with her child being bullied about her appearance:

> *"she described it as she just has to hold it together and she describes herself as being on the edge of like a big black hole and she knows she is right on the edge but she is determined, she also knows that she can't fall into it because if she falls into it then everything will fall apart"*
>
> *Paige, charity worker working with acquired conditions*

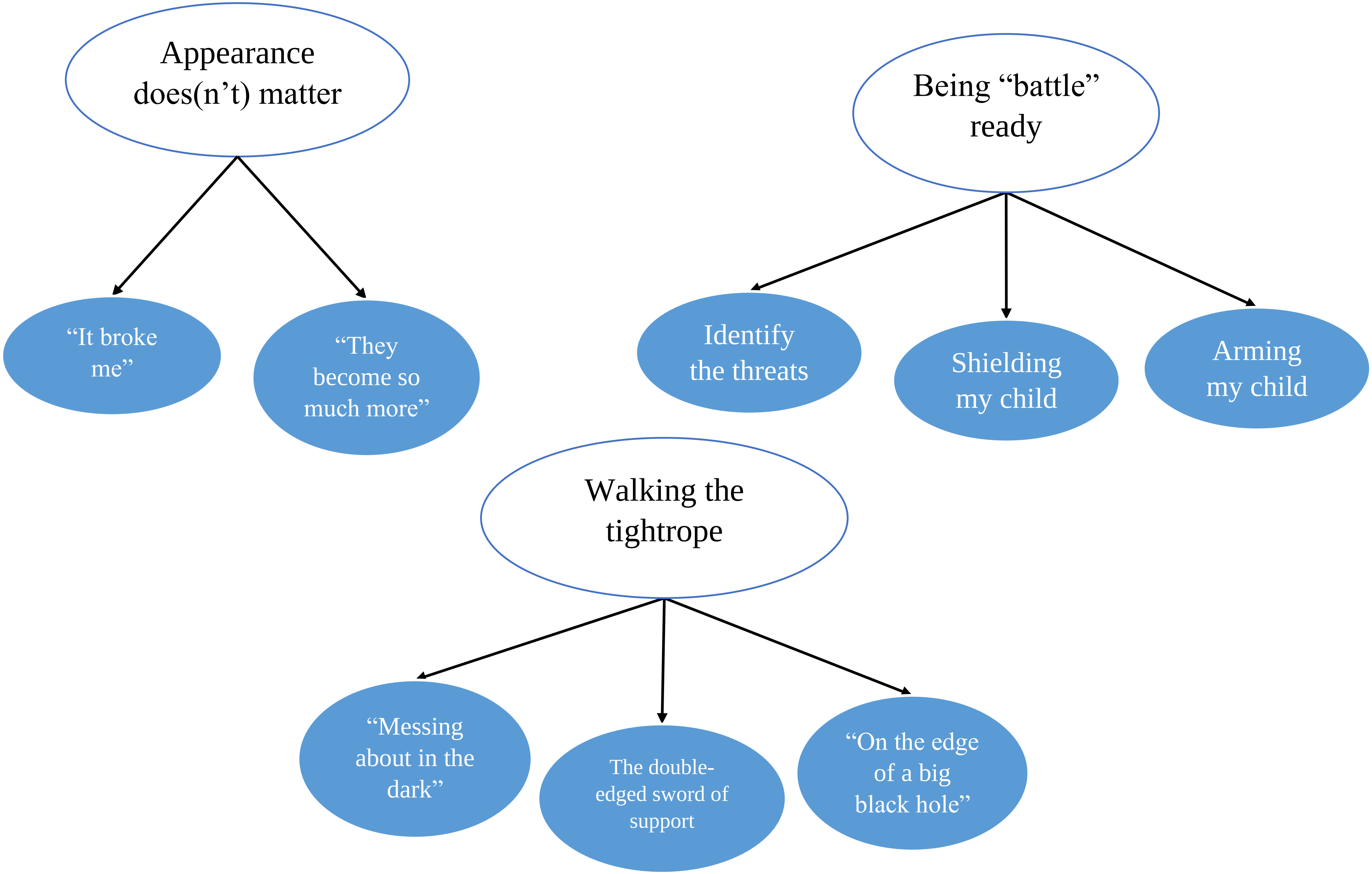

Figure 7: A visual representation of the main themes and subthemes of study 1.

4.4. Discussion

This study aimed to explore cross-condition experiences and support needs of parents of children with a visible difference. Three main cross-condition themes were identified. “Appearance does(n’t) matter”, describes how parents’ attitudes towards the importance of appearance can influence their perception and ability to cope with their child’s visible difference. “Being ‘battle’ ready” describes parents’ concerns about preparing their child to manage these challenges independently. Finally, “Walking the tightrope” describes the balancing act of supporting a child with a visible difference and the lack of confidence and clarity about how to do so successfully. These findings will now be discussed in the context of existing literature and theory.

4.4.1. The psychological impact

Initial reactions of shock and distress to their child’s visible difference were common across parents of children with a variety of conditions and injuries. Consistent with these findings, qualitative and quantitative research with CLP (Costa et al., 2019; Nelson et al., 2012) and burn injuries (Heath et al., 2018a; Hall et al., 2006) have reported shock, distress, and anxiety following their child’s diagnosis or injury. For some parents in this sample, this initial distress was temporary, and they were able to adjust to their child’s condition or injury. For others, psychological distress endured, resulting in difficulties persisting into adolescence. Currently, the existing evidence base does not represent voices of parents with children in mid to late adolescence (Heath et al., 2018a.; Hlongwa & Rispel, 2018; Stock & Rumsey, 2015). The present research therefore builds on current knowledge by increasing understanding of the experiences of parents of older children.

These findings also highlight the common experience of guilt among parents of children with a visible difference, irrespective of their child’s condition, which is a concern. Research indicates these experiences may increase psychosocial distress, for example a study of parents of young children with burn injuries found that feelings of guilt and shame were associated with depressive symptoms and anxiety (Hawkins et al., 2019).

In particular, parents described self-blame associated with the cause of their child's condition or injury (e.g., the injury event or pre-natal development). Feelings of guilt related to causal attributions have previously been reported by parents of young children with CLP and burn injuries (Nelson et al., 2009; Hawkins et al., 2019). In the present study, guilt was reported by both parents whereas previous studies suggest that guilt is particular to mothers. For example, a study investigating causal beliefs in parents of children with CLP found that self-blame had a greater association with anxiety and stress when fathers were excluded from the analysis (Nelson et al., 2009). Authors suggested that this finding was due to the causes of CLP being linked to the mother, due to pre-natal behaviours and events. Previous research into the experience of guilt in this population has also been limited to parents of children under two years old, whereas the present findings build on this understanding by suggesting that guilt persists as children age. The present cross-condition study implies that both parents, of children of any age, might benefit from support to manage feelings of guilt and self-blame

However, as discussed in section 2.2.2. it is important to remember that much of the research about causal beliefs has been conducted in White populations, and that very limited research suggests that some communities from under-represented groups attribute the cause of visible difference to spiritual and religious influences (Olasoji et al., 2007). This understanding of guilt related to causal beliefs may not transfer to all cultural and religious groups.

Although the present research provides an insight into the experience of self-blame, not all parents in this sample felt this way. Existing research has indicated that self-compassion and mindful parenting (parental practices that seek to enhance moment-to-moment awareness) are associated with lower levels of parenting stress (Gouveia et al., 2016). Therefore, increased self-compassion and mindfulness may reduce guilt and self-blame in this population. Further research is needed to better understand the reasons that some parents report experiencing self-blame, whereas others do not. A clearer understanding of the factors contributing to the experience of self-blame could have important implications for guiding clinical practice and support for parents with children of visible differences, as well as other chronic conditions.

4.4.2. Parental perceptions of appearance

Parents' beliefs about the importance of appearance seemed to influence their perception of their child's visible difference. Appearance investment is the importance, meaning, and influence of physical appearance in one's life (Cash et al., 2004). Given the value placed on narrow appearance norms within society (Swami, 2012), it is likely that parents have an idealised image of 'the perfect child'. Consequently, when their child's appearance does not match up to this image, parents may feel they have failed. Existing body image theory and literature suggests that parental attitudes about appearance may also influence their child's experience of body image. The Tripartite model of body image disturbance (Thompson et al., 1999; Abraczinkskas et al., 2012) posits that parents influence the way their children think and feel about their bodies. Existing research has demonstrated that direct parental influence (e.g., appearance-related comments) and modelling of dieting and other appearance-related behaviours, were significantly associated with body image and eating disturbance in both male and female adolescents (Ata et al., 2007; Diedrichs et al., 2016; Emerson & Bogels, 2017; Wood et al., 2007). Investigating parental attitudes toward appearance is important to understand how parents might influence the psychosocial adjustment of young people whose appearance diverges from the norm.

4.4.3. Social learning in the parent-child relationship

Many parents in the present study reported their concerns about the potential impact of their parenting behaviours on their child's adjustment. Paediatric health literature has highlighted the role of the family system and interactions when considering the contributing factors to child psychosocial adjustment (Chernoff et al., 2002) For example, a study of 272 children with asthma and their primary caregivers found evidence for both direct and indirect pathways between parenting emotional expressiveness and child anxiety, depression, and physical health outcomes (Dohnt et al., 2006). This emphasizes a need to reflect on parent-child interactions when exploring adjustment within the family system. Initial findings indicate that interventions which address the importance of the parent-child relationship have also been found to be acceptable to parents (Day et al., 2020). Therefore, it is important to consider the role of the parent-child dyad in future intervention development.

In the present study, parents expressed an awareness that their child may observe and imitate their behaviour. They were anxious to exemplify how to manage appearance-related questions and comments. Theoretical and empirical evidence underpin these concerns. Social Learning Theory claims that children engage in observational learning, wherein they observe, imitate, and model the behaviour of people around them, such as their parents (Bandura & Walters, 1977). Similarly, experimental research has also indicated that children can learn emotions and behaviours, such as phobias, from parents (Gerull & Rapee, 2002). Observations from health professionals in the present study suggested that children imitate their parents' attitudes and approach to their visible difference. Many parents in the current study lacked confidence in their ability to communicate with their child about their visible difference and model adaptive behaviour. Low parenting self-efficacy in parents of children and young people with chronic health conditions has been associated with psychological distress and other health related quality of life measures, such as fatigue (Giallo et al., 2013; Harper et al., 2013). Whereas, parent empowerment has been associated with lower parenting stress, increased parent involvement in decisions, and advocacy for the child in various paediatric healthcare settings (Ashcraft et al., 2019; Bode et al., 2016). Increasing confidence and skills in these areas may protect against distress, whilst also providing parents with the skills to effectively manage their child's condition.

4.4.4. Preparing the child to self-manage

Participants exhibited a deep-rooted drive to protect their child from any threats to their wellbeing. Although protective parental instincts are not a phenomenon unique to this parent group (Chow et al., 2016; Mandell et al., 2005), parents of a child with a visible difference carry additional concerns (e.g., the impact of appearance-related stigma and discrimination). The findings of the present study highlight that this concern can result in further protective behaviours (e.g., attempting to conceal a visible difference, taking a lead in social situations) and anxiety about possible threats (e.g., concerns about or anticipation of bullying or other unwanted attention). Social situations were regarded as particularly threatening, which echoes the existing literature with parents of children with CLP and burn injuries (Hlongwa and Rispel, 2018; Klein et al., 2006). Anxiety about social threats was particularly focused on transition points where children would have to manage independently (e.g., starting secondary school). Given the evidence that appearance and attractiveness are increasingly important in youth culture and appearance-related attributes are highly valued (Benson et al., 1991; Mancuso et al., 2003), it is not surprising that parents of children with visible differences anxiously anticipate their child's increased social independence.

High levels of anxiety are not only distressing for parents, but also a risk factor for overly protective parental behaviours that may negatively impact child development. Quantitative research with parents of children with craniofacial conditions has reported the negative impact that overprotective parenting behaviour can have on children. A study with 36 parents of children with craniofacial conditions found that overprotective behaviour can lead to restrictions in child social interactions, and consequently alienation from support networks. A qualitative exploration of the experiences of 12 parents of burn injured children, found that parents restrict their child's activities (e.g., socialising and participation in hobbies) because they lack confidence in their own and other's abilities to keep them safe from further harm (Horridge et al., 2010). Despite not being directly linked to social threats, this drive to protect can nonetheless impact a child's interaction with the social world. Furthermore, these behaviours may also impact non-affected siblings. Siblings of burn-injured children have reported less involvement in school activities and lower social competence scores when compared with a normative sample (Mancuso et al., 2003). Therefore, it is important to consider the impact of these protective behaviours on social and emotional development of children in the family unit. However, as previously discussed, this existing research on protective behaviour remains limited to parents of children with traumatic conditions or injuries. Therefore, the present findings expand the understanding of protective thoughts, feelings, and behaviours in parents.

Present findings also indicate that parents experience anxiety when considering the challenges of oncoming life stages in their child's life and distress when considering unidentified threats. Barlow's model of Generalised Anxiety Disorder (GAD) conceptualises anxiety as "anxious apprehension" which leads to negative affect characterised by feelings of uncontrollability, and behavioural inhibition (Barlow, 2000). The individual also becomes hyper-vigilant to possible sources of threat and engages in strategies to cope with the anxiety. Coping strategies suggested by this theory include behavioural avoidance and worry in an attempt to problem solve and reduce negative affect (Barlow, 2000). In the present study parents reported worrying about events far into their child's future and framed the consequences of potential threats as highly harmful, whether or not their child had yet encountered the feared event (e.g., bullying). The present study provides insight into social anxiety in parents of children with a range of visible differences and an increased understanding of parent distress related to known and unknown challenges.

Findings highlight different strategies employed to protect their child from appearance-related stigma and discrimination. One approach was to attempt to conceal the visible difference using a variety of methods (e.g., wig use, covering with clothing, or hair styling). This behaviour was discussed more by professionals than parents. One explanation for this difference in accounts, is that professionals may have greater insight into the motivations for these behaviours. Parents may also feel a sense of dissonance and may not want to share this in an interview or focus group discussion. Although concealing a visible difference may reduce parent anxiety about other’s reactions to their child’s appearance, there is evidence to suggest this could be a maladaptive coping strategy. In a seminal study, Moss (2005) identified a significant quadratic relationship between the objective severity of a visible difference, which suggests that individuals are better adjusted if their visible difference is at either end of the severity spectrum (i.e., very severe or not severe) with those in the middle experiencing poorer adjustment. Moss proposed that this is due to the consistency in others’ reactions to the individual’s visible difference at either end of the spectrum. Predictability in the responses of others allows the individual the opportunity to develop coping strategies, However, if the difference is not consistently concealed, individuals and those around them, may not have the opportunity to develop adaptive strategies (Moss, 2005). Therefore, by practicing concealment, parents may inadvertently be missing an opportunity for themselves and their child to develop coping strategies to manage interactions with others.

4.4.5. Accessing support

Some parents felt that healthcare professionals did not adequately address their appearance-related concerns, which in some cases, resulted in distress. Recent qualitative research has identified that healthcare professionals can lack confidence in raising and discussing appearance-related issues and associated distress (Gee et al., 2019). Professionals described not wanting to cause patient harm or make assumptions about the source of the patient’s concern. However, a large proportion of the sample also discussed positive aspects of their management of appearance-related concerns, and the importance of validating feelings and providing reassurance (Gee et al., 2019). This suggests that while professionals want to provide sympathetic care, they (like parents) can lack confidence in how to approach the topic of appearance-related distress. Healthcare satisfaction has been associated with reduced stress, anxiety, depression, and improved quality of life outcomes in parents of children with CLP (Stock et al., 2020). Therefore, it is important that healthcare professionals are aware of challenges faced by parents and can provide support to those who lack confidence. In line with existing literature, these findings provide evidence of a need for further training around appearance-specific support for families and normalisation of appearance talk with parents in healthcare settings.

4.5. Limitations

Whilst including a range of conditions and injuries, the sample remained selective, comprised of parents who were receiving support or engaged with charitable organisations. Some small differences have been reported in preferences for the delivery of psychological support, when comparing samples from charities and hospital sites (Dures et al., 2016). In addition, parents who are involved with charities or currently engaged in social support are more likely to employ effective coping strategies, rather than avoidant coping (Batenburg & Das, 2014). Therefore, this sample may not be transferable to the experiences and support needs of all parents of children with a visible difference. Future work in this area should include parents who are not engaged in support from charitable organizations.

Despite attempting to recruit from a range of backgrounds, it should be noted that the sample was comprised of parents who were mostly highly educated, which can indicate higher socioeconomic status (SES). Sample bias toward higher SES is not uncommon in psychological research (Muthukrishna et al., 2020) but it is important to acknowledge that parents in this sample may have access to resources (e.g., financial support) that could facilitate coping with challenges related to their child’s condition or injury, which individuals from lower SES backgrounds may not. Which again may limit transferability of these findings outside of this sample.

A sample of health and support professional specialist staff with regular contact with this population was purposively recruited to capture the perspectives of professionals working with a range of conditions and injuries. Parents of children with visible differences are likely to have ongoing contact with non-specialist health professionals (e.g., general practitioners, dentists). Condition specific research with non-specialist health professionals found that non-specialists experience challenges in delivering care and support to these families and have a range of unmet training needs (Stock et al., 2020, e.g., lack of confidence engaging in discussions related to their patient's appearance). Future research including non-specialists may provide further insight in how to support these training needs.

Lastly, due to their opportunistic nature, the focus groups were only conducted with parents of children with a single condition (CMN). Due to the social nature of the data collection setting, the findings may not be transferable to all populations and settings. For example, within the focus group discussions, many parents commented that they felt they were among peers and as though they fitted in. As such, they did not speak about feelings of isolation, despite this being a common experience reported by parents of children with other visible differences (e.g., burn injuries, Heath et al., 2018a; eczema, Lewis-Jones, 2006). Nonetheless, these focus groups were valuable because they provided an opportunity to better understand the experiences of parents of children with a rare appearance-affecting condition and contribute to the broader cross-condition themes. Findings from these focus groups also provide a rationale or including parent dyads in future research of this kind.

4.6. Conclusions

Parents of children with a wide range of visible differences experience similar psychosocial difficulties, regardless of the cause or nature of their child's condition or injury and thereby demonstrate commonalities across the experiences of parents of children with a range of visible differences. Parents can be preoccupied with potential future challenges related to their child's visible difference and concerned about how to prepare them to manage these effectively and independently. Some parents felt that when reaching out for support to help their child, their needs were unmet. The experiences described in this chapter highlight a need for further psychosocial support for parents to enable them to manage their own reaction to their child's visible difference, preparing their child to understand, communicate and manage appearance-related challenges independently. Further research, informed by the present findings, needs to examine possible risk and protective factors for distress in these parents, to identify possible psychosocial targets for intervention. These experiences may also be present amongst parents of children with a wider variety of chronic conditions. An understanding of these experiences and further exploration of risk and protective factors would have important implications within the visible difference field, as well as for parents and families of children with health conditions and injuries more broadly.

Chapter five: Mapping qualitative to quantitative design

5.1. Introduction

In line with a sequential exploratory mixed methods design (see section 3.6.1.), study 1 utilised a qualitative design. The aim was to conduct an in-depth exploration of the experiences of parents and identify variables for further investigation in study 2, a quantitative study. One aim of the present study was to investigate whether the qualitative findings generalised to a larger population. A further aim was to investigate whether any of the identified constructs may act as risk or protective factors for psychological distress in parents. The intention of the mixed methods design was to use qualitative data to inform the development and design of quantitative research. This is referred to in the literature as using mixed methods for development (Madey, 1982).

The following section describes how the qualitative themes from study 1 were used to identify pertinent quantifiable psychological constructs for further investigation in study 2. The psychological theory and research evidence that underpins the choice of constructs will also be discussed. A summary table of this mapping process can be seen in Appendix G.

5.2. Mapping themes to psychological constructs

A systematic process of mapping qualitative themes to operationalised psychological constructs was carried out. This process aimed to identify factors that influence parental adjustment to their child's visible difference. Systematic mapping of qualitative to quantitative research has been previously utilised in health research (e.g., Jokiniemi et al., 2018). However, many publications which cite the use of a sequential exploratory model for development do not describe this process in detail. This reduces transparency regarding the rigour of mapping procedures. A lack of clarity around this process also calls into question the aims of the mixed methods, which ultimately threatens the quality of the research conducted. Mixed method theorists have suggested that the initial qualitative findings in a sequential exploratory design should be used to generate theory and hypotheses, which can then be tested through quantitative investigation (Creswell & Plano Clark, 2008). This process was adhered to in the present PhD, followed by a rigorous process of systematic mapping qualitative themes to quantitative data.

The qualitative findings highlighted the complexities of caring for a child with a visible difference. Parents reported many shared experiences which suggest the presence of cross-condition support needs. Mapping cross-condition qualitative themes onto psychological constructs was a challenging process. Parents and professionals described experiences based on their own understanding and insight. A primary challenge was to translate subjective accounts delivered in the participants' own words into operationalised psychological constructs, while ensuring that their experiences were authentically reflected following this shift.

The mapping process and identification of constructs was discussed and reflected upon with the PhD supervisory team. Both the key qualitative findings and the online survey were reviewed by public involvement representatives, who were asked to provide feedback on content (e.g., whether any important topics had been missed). A summary of the qualitative findings was also sent to all participants who took part in the study and the PhD student received positive feedback in response. The student also reflected on their previous clinical experience of working with parents of children with appearance-affecting conditions.

The systematic mapping process was comprised of three phases (See Figure 8). In phase one, the cross-condition themes from study 1 were reviewed. Secondly, existing literature and theory were utilised to identify psychosocial constructs within the qualitative data. This process focussed specifically on identifying factors that influenced adjustment in parents. These phases of the systematic process have been conducted in previous health research which has utilised a similar sequential exploratory mixed methods design (e.g.

Jokiniemi et al., 2018). Finally, measures were selected to assess the identified psychosocial constructs. The following section will describe the cross-condition themes discussed and the theory and literature related to associated psychosocial constructs.

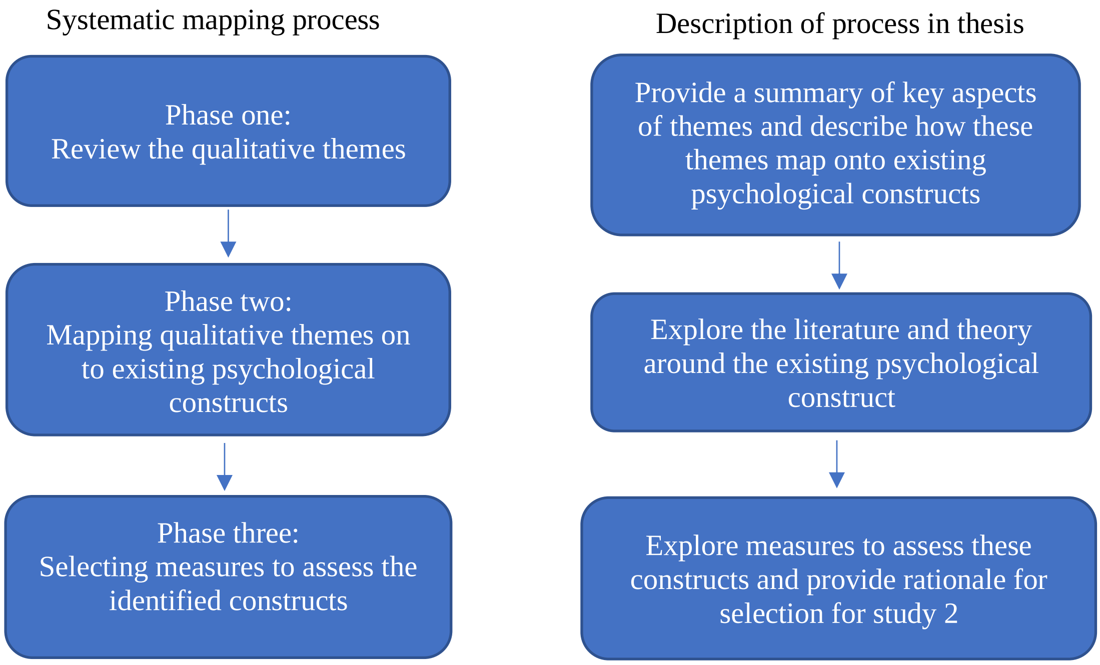

Figure 8: Systematic mapping process on moving from qualitative to quantitative research.

5.3. Parent adjustment: positive and negative affect and parenting stress

As described in the theme ‘Appearance does(n’t) matter”, many parents experienced some degree of psychological distress. Distress was typically contextualised as a response to the initial diagnosis/injury, a reaction to actual or anticipated challenges (e.g., anxiety about bullying or teasing), or concerns about how to provide the right support. Parental psychological distress and stress responses have also been found in the existing condition-specific literature (e.g., Hawkins et al., 2018; Heath et al., 2018a; Nelson et al., 2012). For the purposes of this PhD parental psychological distress and stress were seen as outcomes indicative of poor adjustment.

Some parents also described positive affect (e.g., pride, determination) in response to their child's condition or injury and the challenges it posed. Parent reports indicative of this positive adjustment included a greater acceptance of appearance diversity, empathy for those who are different, and awareness of their own, and their family's resilience. Positive adjustment has also been found in the existing condition-specific literature (e.g. (Eiserman, 2001; Klein et al., 2006). Consequently, parents exhibited aspects of both positive and negative psychosocial adjustment to their child's condition. As discussed previously (section 2.3.1.), this suggests that conceptualisation and measurement of psychosocial adjustment should take a balanced approach that includes both positive and negative cognitions and emotions. The following section explores psychosocial constructs which may influence adjustment in this population.

5.4. Factors associated with parent adjustment

5.4.1. The balancing act of parenting: psychological flexibility and self-compassion

Parents and professionals described the balancing act that parents may struggle with. Parents demonstrated a deep-rooted drive to protect their children from threats to their wellbeing that may result from their appearance difference and felt a responsibility to prepare their children to manage these challenges. However, many lacked confidence in how to best support their child. Specifically, they were concerned about placing too much or not enough emphasis on the visible difference during this process of 'arming' their child. The way parents approached this challenge and their own thoughts and feelings about their performance in this balancing act was central to understanding parent experience.

There were several psychological constructs related to this experience. Parents who appeared to balance concern about their child's visible difference with a desire to set a good model of coping, tended to report reduced distress related to challenging situations associated with their child's visible difference. This ability to live in line with one's values and manage concern or distress mapped onto the construct of psychological flexibility (Hayes et al., 2006).

Psychological flexibility is the ability to orient to the present moment with openness and awareness and behave in accordance with one's values (Hayes et al., 2006). Increased psychological flexibility can promote wellbeing through reducing the compulsion or need for experiential avoidance. Psychological flexibility is fostered through a number of approaches: 1) encouraging individuals to develop an awareness of the negative impact of avoiding or attempting to control feared or unpleasant experiences (including feelings, thoughts, sensations); 2) developing techniques to change the way individuals interact with their thoughts; 3) encouraging non-judgemental contact with psychological and environmental situations as they occur and, 4) helping individuals to identify their values (long-term desired qualities, Hayes et al., 2006) and promote behaviour in line with those actions. The development of psychological flexibility is encouraged through six core processes (Hayes et al., 2006). Psychological flexibility and associated concepts are explored further in section 8.1.1.

Existing research has begun to explore the role of psychological flexibility in supporting wellbeing in individuals with a visible difference. A cross-sectional survey of women affected by lipoedema (progressive fat build-up in the lower parts of the body) found that self-reported psychological flexibility positively predicted quality of life (Dudek et al., 2016). A cross-condition survey with 220 adults with various visible differences found that cognitive fusion partially mediated the relationship between body evaluation and unhelpful body image coping strategies (e.g., appearance-fixing behaviours), and experiential avoidance partially mediated appearance-related behavioural avoidance (Zucchelli et al., 2020). Therefore, psychological flexibility seems to be a key mechanism in the wellbeing and coping strategies employed by individuals with visible differences. However, no work has examined psychological flexibility in parents of children with visible differences and whether this could be protective against psychological distress.

As described in the theme “Walking the tightrope”, parents who were invested in doing the ‘right’ thing for their child (e.g., setting a good example of coping, preparing them to manage challenges), also had high expectations of themselves. They bore the burden of responsibility for preparing their child to manage appearance-related challenges. They often blamed themselves if/when their child struggled. This burden of responsibility and self-blame sometimes resulted in parents feeling stressed or anxious, or low if their child struggled. This experience could be mapped on to the construct of self-compassion (being kind and understanding toward oneself in instances of pain or failure, Neff et al., 2007).

Self-compassion includes the following concepts: 1) being kind and understanding toward oneself rather than being self-critical, 2) perceiving one’s fallibility as part of the human condition and experience rather than as isolating and, 3) holding painful feelings and thoughts in mindful awareness rather than avoiding or over identifying with them (Neff, 2003). Barnard & Curry (2011) have proposed that these three components of self-compassion are related to each other, and one component can strengthen the others. For example, if an individual is caring and understanding towards themselves, they may be less likely to feel shame about any perceived imperfections (Brown, 1999). Rather than withdrawing from relationships and believing they struggle with failures alone, they may be more likely to share their struggles or observe that others have similar difficulties. Self-kindness might also allow the person to stay in the present and adopt a balanced view, whereas self-judgement focuses on the past or anticipation of future difficulties (Barnard & Curry, 2011b).

The benefits of self-compassion have been widely explored within the health psychology literature. A meta-analysis of 15 studies found that self-compassion was positively associated with the self-reported practice of health-promoting behaviours (eating habits, exercise, sleep behaviours, and stress management; Sirois et al., 2015). Authors concluded that this effect was in part due to its association with adaptive emotions, such as positive affect. In a study with adults (n=287) and adolescents (n=235), maternal support and family functioning have been identified as predictors of individual differences in self-compassion (Neff & McGehee, 2010). Self-compassion was also found to partially mediate the link between family functioning and wellbeing. This indicates that self-compassion may have an important role in wellbeing within the family.

Self-compassion and mindfulness have also been associated with improved well-being in parents. Mindful parenting has been described as a set of parental practices that seek to enhance moment-to-moment awareness in the parent-child relationship (Gouveia et al., 2016). A questionnaire study with 333 parents found that higher levels of self-compassion and dispositional mindfulness were associated with higher levels of mindful parenting, which was also associated with lower levels of parenting stress (Gouveia et al., 2016). Interventions aiming to increase self-compassion have also been utilised to improve parent wellbeing. A randomised controlled design study found that mothers who underwent an 8-week mindfulness-based intervention exhibited significantly less anxiety, stress, and psychological distress, when compared to those in the control group (Perez-Blasco et al., 2013). These results suggest that interventions to increase self-compassion can be beneficial in reducing psychological distress in a parent population.

Self-compassion interventions have also been found to be effective in reducing body dissatisfaction. One study found that women who took part in a brief 3-week self-compassion intervention experienced significantly greater reductions in body dissatisfaction, body shame, and appearance-related contingent self-worth (self-esteem based on social approval or social comparisons, Crocker & Wolfe, 2001), as well as increased self-compassion and body appreciation, when compared to a control group (Albertson et al., 2015). A self-compassion intervention has also been tested in a visible difference population. In a sample of 50 participants with a range of visible skin conditions (e.g. acne, eczema, psoriasis) who experienced body image distress, those allocated to a self-compassion writing intervention had significantly improved self-compassion and reduced negative affect, compared to the control group (Sherman et al., 2019).

One example of a therapeutic intervention which aims to promote psychological flexibility and self-compassion is Acceptance and Commitment Therapy (ACT; Hayes et al., 2006). ACT teaches skills to manage difficult thoughts and feelings by engaging in the six-core processes described in section 8.1.2.2. Self-compassion is also an integral aspect of ACT and has theoretical and empirical associations with psychological flexibility (Luoma & Platt, 2015; Marshall & Brockman, 2016, see section 6.4.3. for greater detail).

Psychological flexibility and self-compassion are important constructs for coping in both the general parent population and individuals with appearance concerns. However, the role of psychological flexibility and self-compassion in parents of children with visible differences has not been examined via research. Study 2 explored whether there is any association between parental psychological flexibility and self-compassion and parent adjustment.

5.4.2. Getting it “right”: parenting self-efficacy and knowledge

Parents described that they lacked confidence in their abilities to support their child and prepare them to cope with challenges related to their visible difference. This lack of confidence often resulted in parents feeling worried that they were “ill-equipped” to support their child. Parents believed that the approach they adopted when supporting their child would be influential in shaping their child’s ability to independently manage appearance-related challenges. This data mapped onto the psychological construct of perceived self-efficacy. When parents expressed that they lacked knowledge or skills, they also described feeling fearful for their child’s future.

Perceived self-efficacy is defined as an individual’s beliefs about their capabilities to produce levels of performance that exercise influence over events that affect their lives (Bandura, 1994). Beliefs about one’s self-efficacy determine how people feel, think, motivate themselves, and behave. These beliefs produce behavioural outcomes stemming from cognitive, motivational, and affective processes (Bandura, 1994). High self-efficacy appears to enhance feelings of accomplishment and well-being. Individuals who feel confident in their capabilities approach tasks or experiences as challenges to be mastered, rather than threats to overcome (Bandura, 1994). These individuals also recover their sense of self-efficacy quickly after failures or setbacks.

The impact of self-efficacy on health and well-being has been widely explored.. In a review of 204 experimental studies, it was identified that self-efficacy has a causal effect on health-intentions and behaviour (Sheeran et al., 2016). Self-efficacy in parents of children with chronic illnesses has also been associated with increased parental and child well-being. A cross-sectional survey study with 32 parent-children pairs found that self-efficacy and self-management scores were associated with health-related quality of life (Bravo et al., 2020). These authors concluded that caregivers who are providing health care and management need ongoing education and support. This is important to note as parents of children with visible differences may be providing high levels of care at home (e.g., eczema and burn injury treatment regimens). In addition, Bravo et al found that child and parent self-management training could lead to a decrease in the use of limited healthcare resources. In study 1, parents of children with visible differences expressed concern about how to manage a variety of issues (e.g., communicating with their child about their visible difference, promoting confidence and resilience in their child). The present literature reinforces the importance of equipping parents with self-management skills, for both the family and efficiency of healthcare service use more broadly.

Interventions aimed at increasing self-efficacy have been utilised to improve child outcomes. A study with 45 mothers and 44 fathers examined the effectiveness of parent management training to increase self-efficacy in parents of children with Asperger syndrome (Sofronoff & Farbotko, 2002). When compared to the control group, parents in the intervention group reported fewer problem behaviours in their child and greater parental self-efficacy. This suggests that self-efficacy interventions may help parents to manage difficult behaviours in their children, which may reduce stress or burden of care for parents.

In a study assessing the effectiveness of an online self-efficacy intervention for parents of children with type 1 Diabetes, a significant increase was found in pre- and post-intervention parent self-efficacy scores (Merkel & Wright, 2012). However, Merkel and Wright (2012) do not expand on parent outcomes beyond the increase in self-efficacy. From this it can be concluded that these interventions may have utility for increasing self-efficacy in these parent population, but the impact on broader parent wellbeing remains unknown.

Some parents struggled when they felt they had insufficient information about their child's treatment or condition to support them. Parents often engaged in information seeking behaviours, both from healthcare professionals and parent peers. These individuals reported mixed experiences when reaching out for support and information. Those who felt their information and support needs were not met, expressed distress.

Self-efficacy and parent knowledge share some characteristics with the concept of patient activation (patients having the knowledge skills and confidence to manage one's health, Hibbard et al., 2004). The patient activation literature has proposed that patients play a part in shaping the need for and outcomes of care (Greene & Hibbard, 2011). Patient activation is informed by the Chronic Illness Care Model (Bodenheimer et al., 2002) which emphasises patient-oriented care, with patients and families integrated as members of the care team. Within this model it is essential that individuals are "activated" and have the skills, knowledge, and motivation to be involved in decision-making (Hibbard et al., 2004). Cross-sectional research has found that patient activation is strongly associated with a wide range of positive health-related outcomes (Greene & Hibbard, 2012). In contrast, a randomised controlled trial found that patient involvement schemes, such as patient feedback, have not improved mental health outcomes (Rise et al., 2016). However, this could be due to a lack of consistent patient integration throughout the course of treatment. These findings suggest that self-efficacy and self-activation require that patients and family be involved at all stages of the care process to achieve knowledge, skills, and motivation to improve health outcomes.

Self-efficacy has been widely linked to increased wellbeing and quality of life, however evidence regarding parental outcomes is lacking. Therefore, the second study of the PhD explored whether there is an association between parent self-efficacy and parent adjustment and parent-reported knowledge and adjustment.

5.4.3. Parent-child communication

Parents consistently described concerns about communicating effectively with their child about their visible difference. They were worried about how to pass on information about the condition/injury and manage questions or conversations instigated by the child. Parents were also conscious that they wanted to foster an attitude of acceptance and positivity about appearance diversity. They hoped that educating and modelling open communication would help their child to develop an understanding and positive narrative about their own visible difference. Individuals who reported anxieties about this process, discussed the negative impact this had on their wellbeing.

Parent-child communication has been found to be an important contributing factor to health outcomes in families with a child with a chronic health condition. In a study of 217 families of a child with type 1 diabetes, it was found that positive family communication predicted greater treatment adherence and glycaemic control (Iskander et al., 2015). Effective communication appears to be an important process in the management of long-term conditions. Based on the existing literature and the findings of study 1, parental confidence in their ability to communicate about their child’s visible difference was deemed to be an area that warranted further investigation in the second study of this PhD.

5.4.4. Optimism

When parents spoke about their experiences, some demonstrated a more positive life orientation or outlook than others (also known as optimism, Carver et al., 2010). These parents tended to be more orientated towards the attitude that their child’s visible difference was not a problem or, in some cases, highlighted the positives of their child having a visible difference. A few of these parents also spoke about their hope for their child’s and their family’s future. As a result of this positive orientation, these parents appeared to experience less distress related to their child’s visible difference.

Optimism is an individual difference which reflects the extent to which people hold “generalised favourable expectancies” for the future (Carver et al., 2010). Simply, increased optimism is associated with better subjective well-being during adversity and is related to reduced feelings of hopelessness, a risk factor for depression (Alloy et al., 2006). The trait of optimism may provide cognitive, coping, and contextual resources (e.g., social integration) that promote well-being (Carver et al., 2010).

The theoretical underpinning of optimism is linked to expectancy-value models of motivation, which assume that behaviour reflects the pursuit of goals (desired states or actions; Carver et al., 2010). The more important the goal is to the individual, the greater the value (Higgins, 2006). In motivational models, expectancy is the confidence that the goal can be attained (Carver et al., 2010). If an individual lacks confidence in their ability to obtain the goal, they may reduce or withdraw their effort towards it, whereas individuals who are confident in their ability to obtain the goal persevere, even in times of difficulty. This has important implications for how people deal with challenging situations (Carver et al., 2010). Optimists tend to expect good outcomes, even when life is difficult, whilst pessimists tend to expect poor outcomes, which can lead to increased negative feelings such as anxiety, anger and sadness (Scheier & Carver, 1998; Scheier & Carver, 1992).

The relationship between optimism and psychological well-being has been explored in parents of children with chronic conditions. A cross-sectional survey study with 100 parents of children with cancer found optimism to be significantly associated with increased life satisfaction, and reduced anxiety and depression (Fotiadou et al., 2008). Furthermore, positive life orientation has been associated with increased psychological wellbeing in parents of children with appearance-affecting conditions. A cross-sectional survey study with parents of children with CLP found that positive life orientation was found to be protective against psychological distress for both mothers and fathers (Stock et al., 2020). However, it is important to acknowledge that the research described is cross-sectional and does not examine how life orientation may change over time. However, it is possible that optimism may have a role in supporting wellbeing in parents of children with a wider range of visible differences.

There is also evidence to suggest that psychological interventions aimed at increasing optimism can be effective. A meta-analysis of 29 studies evaluating the effectiveness of interventions in a range of populations (Malouff & Schutte, 2017) concluded that interventions that included the “Best Possible Self” programme had the highest weighted mean effect sizes for increased optimism, when compared to other methods. The “Best Possible Self” intervention involves participants developing goals for visualising a best positive future self (Meevissen et al., 2011). Interventions to increase optimism have also been designed specifically for parents. One such intervention is the Positive Family Intervention (PFI), developed to help families manage child behavioural difficulties (Durand et al., 2009). Utilising a cognitive-behavioural approach, PFI aims to provide family members with coping skills. However, evaluations of this intervention focus on the child outcomes rather than those related to the wellbeing of parent participants.

There is evidence to suggest that optimism is related to psychological wellbeing, particularly during challenging times. Parents of children with visible differences experience a multitude of challenges throughout their child’s life. The condition specific literature has indicated that optimism is related to parental wellbeing, however further exploration of the role of optimism is warranted in a cross-condition investigation. The second study explored whether there is an association between optimism and parent adjustment.

5.4.5. Social situations

Parental anxieties were primarily focussed on social challenges related to their child’s appearance difference (e.g., bullying, or social exclusion). This included concerns about children socialising within their own circles (e.g., at school or with friends), as well as interacting with the public. The possible impact of living with a socially stigmatised condition or appearance is addressed in detail in section 2.2.1.

Professionals also shared examples of parents who had expressed concerns about the potential negative social impact of their child's appearance. Professionals emphasised that these experiences or fears can underpin parents' drive to seek appearance altering treatment. These experiences were described in the sub-theme, "Identify the threats". Parent perceptions of the degree of social challenges experienced by their child could provide insight into how significant parents believe this threat to be. Given parents' concerns about the impact of these difficulties, the perceived severity of social challenges may be a predictor of parent distress.

Parents specifically reported anxieties related to their own responses to unwanted attention and reactions of others. In addition, in the sub-theme "Arming my child", parents discussed the desire to set a good example for their children when interacting with others (e.g., answering questions or addressing appearance-related comments). Parents who spoke about feeling confident in these interactions reported that their children were also more confident. The parents themselves also reported less distress. This suggests that parent social confidence in these situations could be beneficial for both parent and child wellbeing.

5.4.6. Perceived social support

All parent participants involved in study 1 were recruited through charitable organisations. Consequently, they are more likely to be engaged in some form of social support than families who are not connected with a charity. Many parents also discussed sources and experiences of social support during the interviews and focus groups. Parents who perceived that they had been supported through challenges related to their child's visible difference spoke about this being an important resource in helping them to cope. For example, parents who spoke of supportive friends and family appeared less concerned about social challenges from people close to them.

The role of social support as a coping resource has long been of interest to psychologists (Zimet et al., 1988). Social support has been described as an "exchange of resources between at least two individuals perceived by the provider or the recipient, to be intended to enhance the wellbeing of the recipient" (Shumaker & Brownell, 1984, p. 11). However, it has also been proposed that the resources provided by others can have either a negative or positive effect (Cohen & Syme, 1985). The subjective-objective dimension of social support has also been highlighted by some authors. Lin (1986) reported that social support can be perceived or actual and can be either instrumental (e.g., tangible, practical support) or expressive (e.g., social networks and confiding partners).

The mechanism by which social support acts as a coping resource has also been considered (Zimet et al., 1988). Firstly, there is evidence to support the hypothesis that social support may be directly helpful, regardless of the magnitude of the stress in an individual's life (Broadhead et al., 1983). Whereas it has also been argued that social support acts predominantly as a buffer by protecting individuals from the harmful impact of stress (Lazarus & Folkman, 1984; Penninx et al., 1997). It has also been proposed that social support enhances self-esteem and sense of control, which leads to a more positive emotional experience (Pearlin, 1999). Cohen and Syme (1985) suggest that the focus of social support is to aid in the maintenance of health and disease recovery. One possible mechanism posited is that by increasing self-esteem and positive feelings, social support may indirectly strengthen the immune system and reduce recovery time from illness and susceptibility to disease (Cohen & Syme, 1985).

The existing condition-specific literature has highlighted that social support is likely to be an important coping resource for parents of children with a visible difference. In a cross-sectional study of 103 parents of children with CLP, higher levels of social support were predictive of less family impact, lower psychological distress, and greater positive adjustment to having a child with a craniofacial condition (Baker et al., 2009). Bogart et al's (2017) mixed methods study that explored the experiences of attending a support conference with individuals with Moebius syndrome (n=50) and parents of children with Moebius syndrome (n=57), reported that parents find practical social support (e.g., advice from peers and professionals) most beneficial. Therefore, this indicates that parents of children with a visible difference can perceive instrumental social support to be an important coping resource.

Existing research also suggests that parents of children with a visible difference may lack adequate social support. Parents of children with visible differences have reported feelings of isolation. A qualitative interview study of caregivers of burn-injured children found that parents reported experiences of psychological isolation and feeling alone in their experiences (Heath et al., 2018a). These individuals also spoke about a sense of physical isolation, particularly in the initial period after injury when their child was hospitalised. Consequently, despite social support being an important tool for coping and adjustment, some parents of children with visible differences may experience isolation and a perceived lack of social support from others.

Social support can be beneficial for parents of children with visible differences. However, there is also some indication within the literature that there may be an unmet need for social support in this population. The findings related to social support in parents is currently restricted to condition-specific research. Consequently, we cannot make generalisations about whether social support is a protective factor against distress in parents of children across conditions and injuries. The second study of the PhD investigated whether there is a relationship between perceived social support and adjustment in parents of children with a range of conditions.

5.4.7. Parental perceptions of appearance

When parents reflected on the impact of their child’s visible difference, their responses seemed influenced by the degree to which they themselves valued appearance as an attribute. Parents discussed how much their child’s visible difference concerned them and whether they felt that appearance was an important aspect of their child’s identity. Health and support professionals observed that some parents communicated greater concern and preoccupation with the aesthetic aspects of the condition or injury (e.g., scarring) than others. These individuals seemed to experience greater distress than those who were less focussed on appearance. Parents’ differing communication and attitudes towards their child’s appearance is presented in the theme “Appearance does(n’t) matter”. These data may reflect the varying degrees of parental appearance investment (the extent to which one’s appearance is integral to self-worth; Cash & Pruzinsky, 2004).

The degree to which parents valued appearance as an attribute was sometimes reflected in the way they managed their reactions to their child’s unusual appearance. Concealing their child’s appearance difference (e.g., using wigs to cover hair loss in alopecia) was a strategy employed by some parents. The use of these appearance-focussed coping strategies suggested that parents recognised and reaffirmed the belief that we live in a world where an attractive appearance is highly valued. Associations have been found between psychological **in**flexibility (acting in line with thoughts and feelings rather than being guided by values) and negative body image coping behaviours (e.g., appearance fixing) in both the general population and a visible difference population (Mancuso, 2016; Zucchelli et al., 2020). These findings have indicated that psychological flexibility processes mediate the relationship between body image evaluation and unhelpful body image coping strategies. The impact of carrying out appearance-fixing behaviours on a close other (e.g., a parent carrying out these behaviours on their child) has not been explored in research. Therefore, the second study of this PhD took a novel approach to explore whether conducting appearance fixing behaviours was associated with parental psychological distress related to their child.

5.5. Reflexive analysis of the mapping process

Following this systematic process of mapping was difficult and challenged me to closely examine the qualitative data and consider the overall aims and research questions of the PhD. The PhD primarily aimed to explore cross-condition experiences of parents of children with a range of appearance-affecting conditions and injuries. Namely, the themes that were constructed in study 1, and then considered for further investigation in study 2, were required to occur across conditions and injuries. This was to provide an evidence base for the commonalities in the experiences of parents of children with different visible differences. There were difficulties that parents highlighted based on the specific symptomology of their child's condition or injury. For example, parents of children with a significant facial visible difference, found bonding with their child challenging due to communication difficulties (e.g., facial expressions). There were many more similarities between parent reports than there were specific incidences such as this, but some of these were difficult to let go of, as they represented a significant challenge experienced by a small number of parents.

Once the cross-condition themes had been identified, it was also difficult to choose which constructs would be taken forward to study 2 for further exploration. Another aim of the PhD was to investigate experiences and protective and risk factors for adjustment in this population, with the view to identifying unmet support needs and developing support. Consequently, a pragmatic decision was made to focus on constructs that were more amenable to intervention. As a result, some observations from the data of study 1 were not investigated in study 2. Parenting styles (e.g., authoritative, authoritarian, and permissive) which were discussed in section 2.5.1., were considered to be difficult to effectively measure and target through intervention.

Overall, the mapping process required me to reflect upon my understanding of the data and psychological constructs identified. This required a rigorous interrogation of the data and my own research decision making. It was also necessary for decisions to be made in the context of wider theory and evidence around these constructs. As I became more familiar with the relevant literature, I grew in confidence that I was accurately representing the experiences described in study 1.

5.6. Chapter summary

This chapter provided a discussion on the process of mapping qualitative data onto measurable psychosocial constructs. The following constructs were identified in the qualitative data: psychology flexibility, self-compassion, self-efficacy, parent-child communication, optimism, parent social confidence, parent-reported social challenges, social support, appearance investment, appearance fixing behaviours, and parent knowledge of condition/injury. Each psychosocial construct was described, and the relevant existing literature and theory explored. The measures selected to measure these constructs are described in the following chapter, in relation to the survey study design. The following chapter will also address the design and conduct of a mixed methods online survey which aimed to increase generalisability of the qualitative findings and explore risk and protective factors for parent psychological distress.

Chapter six: Study 2, an online survey exploring the risk and protective factors for psychological distress and stress in parents of children with a visible difference.

6.1. Introduction

The qualitative findings of study 1 suggested that parents report similar experiences (referred to as cross-condition experiences), regardless of the nature or cause of their child's visible difference. A cross-sectional survey design was then selected as an appropriate method to investigate potential associations between the outcome variables of parental affect (the experience of positive emotions, such as excitement or pleasurable engagement and negative emotions, such as guilt or anxiety; Watson & Clark, 1988) and stress (the emotional and cognitive response when individuals perceive that they cannot cope with demands being made on them or threats to their wellbeing; Lazarus, 1966), and various psychosocial constructs described by the participants of study 1. The rationale for the use of a sequential exploratory mixed methods design has previously been described in detail (see section 3.6.1.).

Cross-condition experiences identified in study 1, were used to identify pertinent quantifiable psychological constructs, which could be measured and assessed in the present study. This systematic process of mapping qualitative themes onto quantitative measures, by drawing on existing literature and theory from relevant fields of expertise (e.g., visible difference, parenting, paediatric health, body image), is outlined in detail in the previous chapter. Based on this review of the literature, it was proposed that these identified constructs may also act as potential risk or protective factors for parental affect and stress.

The core constructs that identified were: psychological flexibility (Hayes et al., 2006); self-compassion (e.g. Gouveia et al, 2016; Albertson et al., 2015); perceived parenting self-efficacy (e.g. Bravo et al., 2020); perceived social support (e.g. Bogart et al., 2017); social confidence in response to social stigma (e.g. Hlongwa and Rispel, 2018);

optimism (Stock et al., 2020); parent-child communication (Iskander et al., 2015), and appearance investment (Cash and Pruzinsky, 2004).

Other variables relating to the nature of the child's visible difference and the parents' perceptions of challenges faced by their child were also identified. These were: age of their child; the type of condition or injury that resulted in their child's visible difference; parent's perceptions of noticeability of the appearance difference and whether the parent perceived that their child was experiencing social challenges related to their visible difference (e.g., teasing), and the emotional impact of this. Finally, parent-reported knowledge of their child's condition and satisfaction with care were also identified as being potentially important in understanding parents' experiences.

The selection of these variables was informed by the findings of study 1 and the existing visible difference and paediatric health literature (see section 5.3. and 5.4.). Additionally, these variables align with Belsky's model of the determinants of parenting (Belsky, 1984). This theory posits that parent functioning and child adjustment can be attributed to three domains: parental psychological resources, characteristics of the child, and contextual sources of stress and support. Belsky's model provides a clear rationale for investigating parenting affect and stress as an outcome to indicate parent adjustment. The predictor variables identified also represent various other aspects of contextual stress and support (e.g., perceived social challenges, perceived noticeability of the appearance difference, age of the child, perceived social support) and parent psychological resources (e.g., self-compassion, psychological flexibility, self-efficacy). Therefore, the quantitative design selected to assess adjustment is grounded in health and parenting evidence and theory.

The following chapter will outline the second study of this PhD and discuss the findings, with reference to existing literature and theory. The aim and research questions for the present study were as follows:

Research question and aims

Research question:

- Which psychosocial factors are associated with positive and negative affect and stress in parents of children and young people with appearance-affecting conditions and injuries?

Aims:

1. To increase the generalisability of the findings from the qualitative data collected in study 1 of the PhD
2. To investigate whether potential risk and protective factors (identified in study 1) are associated with parent affect and stress
3. To identify possible targets for psychosocial intervention

Hypotheses:

The following hypotheses were informed by the findings of study 1 (sections 4.3., 4.4. and 5.2.), existing visible difference and health literature and theory (see sections 5.3. and 5.4., and parenting theory (see sections 2.5. and 6.1.).

1. *Given the cross-condition themes generated in study 1, there will be no significant difference between congenital and acquired appearance-affecting conditions or injuries, on any of the four outcomes: negative affect, stress frequency, stress difficulty or positive affect.*
2. *Based on the findings of study 1 and existing visible difference, health, and parenting literature and theory, noticeability of the visible difference to parents, noticeability to others, parent-reported teasing, appearance investment, and parent-report appearance-fixing behaviours will be significantly positively associated with negative*

affect, stress frequency, stress difficulty, and significantly negatively associated with positive affect.

3. *Based on the findings of study 1 and existing visible difference, health, and parenting literature and theory, parental self-compassion, parent psychological flexibility, effective parent-child communication, parent-reported knowledge of their child's condition/injury, parent self-efficacy, perceived social support, and parent optimism will be significantly negatively associated with negative affect, stress frequency, stress difficulty, and significantly positively associated with positive affect.*
4. *Based on the findings of study 1 and parenting theory, child age will be significantly negatively associated with negative affect, stress frequency, stress difficulty, and significantly positively associated with positive affect.*
5. *Based on the findings of study 1, a parent of a child who identifies as a girl will be significantly positively associated with negative affect and significantly negatively associated with positive affect.*
6. *Based on the findings of study 1 and existing visible difference literature, parent gender will significantly predict parent affective and stress outcomes.*

6.2. Method

6.2.1. Study design

Data were collected using an online survey design comprised of standardised psychosocial measures and study specific measures. An online survey design also allowed for a large and heterogeneous sample of parents and carers (e.g., a large range of conditions, different family structures etc.) which was important to increase the generalisability of the findings and allow for more robust conclusions about the support needs of parents and carers, and thus provide direction for intervention development. Qualitative data were also collected to enhance and clarify the quantitative data and give participants an opportunity to elaborate on their experiences in their own words (Greene, Caracelli & Graham, 1989). As qualitative data were being used for complimentary purposes, the quantitative and qualitative data were mixed using a thread approach (Moran-Ellis et al., 2006; described in section 6.2.7.6.). This use of sequential mixed methods is in line with the Medical Research Council guidelines for the development of complex interventions (Skivington, 2021) which emphasises the importance of identifying an evidence base in the development stage of intervention design.

6.2.2. Survey design

This section will describe the process of measure selection for the online survey of the present study. The full survey can be seen in Appendix H.

6.2.2.1. Measure selection

Several factors were considered when selecting measures to assess constructs identified in study 1, including the psychometric properties of candidate measures, and prevalence of their use in the field. Standardised measures have the advantage of having established and published reliability and validity statistics. Where possible and appropriate, measures were chosen that had been previously utilised in populations close to that investigated by the present PhD (e.g., parents of children with chronic conditions, parents in the wider population) and had published figures which indicated good reliability and validity. However, it is worth noting that a lack of consensus in measurement in research with visible difference populations has been acknowledged in the existing literature (Stock et al., 2016). Standardised measures vary in scoring and cut off points, and this variability in reporting can influence the findings and conclusions drawn from a study (Due et al., 2005). Research studies are more comparable if the same measures have been used, which enables researchers to reliably draw conclusions across a body of work with a specific population, allowing for this work to be enhanced and built upon by future research. Thus, when selecting measures their frequency of use in the field of visible difference was considered.

Participant experience of completing the survey was also important, particularly in relation to the length of the chosen measures and whether completion would cause fatigue. Response burden in participants has been discussed at length in health research (Rolstad et al., 2011). Some authors have proposed that increased response burden could lower response and completion rates, and reduce data quality (Diehr et al., 2005; Snyder et al., 2007). Although, a meta-analysis of 20 studies found an association between response rate and questionnaire length, it was unable to conclude that number of items was the only influencing factor (Rolstad et al., 2011), suggesting that the impact of questionnaire content also plays a role. To ensure that the content of the current questionnaire was relevant and acceptable to this population, PPI input was sought prior to data collection (See section 6.2.3.).

It was important to ensure that all items within the chosen measures were relevant to the specific appearance-related challenges and experiences of parents of children with a range of visible differences/injuries; this proved challenging. The target group is also relatively small and currently under-researched. As a result, very few measures have been designed or validated for this population, and of those that have, most are limited to condition-specific research (e.g., CARe Burn scales, Griffiths et al., 2021). Thus, some candidate measures were rejected for failing to reflect the specific experiences and challenges of these families (see section 6.2.2.). From an ethical point of view, it did not feel appropriate to use these measures to collect data that might poorly reflect their experience and potentially impact the validity of the findings. As a result, the PhD student either constructed study specific items to measure certain constructs (e.g., parent self-efficacy) or adapted measures to meet the needs of the cross-condition parent population (e.g., CEN-Q; treatment knowledge and satisfaction with care). The content of the items developed by the

PhD student was informed by the findings from the first study and all survey items were reviewed by PPI members to maximise their face validity.

The measures included in the survey are outlined below. All scales used in the present study had Cronbach's alpha scores of > 0.68, with all but one scale (Adapted CEN-Q; knowledge and treatment satisfaction) being α > 0.7, indicating good internal reliability (Ponterotto & Ruckdeschel, 2007).

Parent positive and negative affect

The Positive and Negative Affect Scale (PANAS) represents the two dominant dimensions of affective structure that have been identified in psychological research (Watson & Tellegen, 1985). Comprised of two 10-item mood sub-scales (20 items total), the items are a list of feelings and emotions (e.g., guilty, scared, excited, determined etc.). Respondents indicate on a 5-point Likert scale of very slightly or not at all (1) to extremely (5), how much they have been feeling that emotion over the last week. Both PANAS sub-scales demonstrated good internal consistency in the present sample (positive affect, α = 0.91, negative affect, α = 0.88) and good test re-test reliability over eight weeks in previous research (positive affect, r = 0.47 – 0.68, negative affect, r = 0.39-0.71, Watson, Clark, & Tellegen, 1988). This scale was used as a measure of parent adjustment to their child's visible difference. The PANAS has been widely used with samples of parents in wider paediatric literature. For example, in a study of emotional expression among 69 parents of children with serious illness (e.g., neuromuscular conditions, childhood cancer; Hexem et al., 2013).

As the PANAS has not been used in visible difference research before, which may consequently limit comparisons that can be drawn between this and other studies in the field, the Hospital Anxiety and Depression Scale (HADS, Zigmond & Snaith), was considered, due to its prevalence in clinical research settings). However, the HADS has been criticised for variance in its sensitivity and specificity when measuring anxiety and depression

(Mitchell et al., 2010). It is possible that the PANAS has not been as commonly used as other measures of negative affect, due to the focus of traditional clinical psychology to detect negative emotions and treat them as symptoms of psychological disorders (Schlechter et al., 2019). The PANAS was ultimately selected because its items reflect *both* positive and negative emotions, which represents parent experiences described in study 1. It also has items that accurately reflect words used by parents in the interviews and focus groups to describe their emotions (e.g., guilty, scared, proud, and determined).

Parenting stress

The brief Paediatric Inventory for Parents (PIP) is a measure of parenting stress specifically related to caring for a child with an illness (Streisand et al., 2001). This measure, previously used with carers of paediatric patients with chronic illness, is a 12-item scale divided into four subscales: communication, medical care, role function and emotional functioning (Casaña-Granell et al., 2018). Each item receives two scores: one for the frequency of the stress (e.g., "How frequently do you worry about long-term consequences of the condition or injury?") and one for the stress difficulty (e.g., "How difficult do you find it to deal with worrying about the long-term consequences of the condition or injury"). Only the emotional functioning subscale (3 items) was chosen to measure stress in the present study, because the other subscales focussed on the stress of the child being in hospital and receiving active treatment (e.g., "Helping my child with medical procedures"). These items may not be as relevant for parents of children who are less likely to have regular ongoing medical treatment regimens (e.g., alopecia, limb difference etc.). It demonstrated good internal consistency in this sample on both the frequency and difficulty dimensions (frequency $\alpha = 0.85$, difficulty $\alpha = 0.89$). This subscale was selected because it reflected the parent-reported experience of stress from the study 1.

As the PIP is also not widely used, which may hinder comparisons to other research, the Parenting Stress Index (PSI, Abidin, 1995) which has more widespread use was also considered. Although popular in parenting research, the PSI was not specifically designed for parents of children with health conditions. Consequently, many of the items appeared irrelevant or unfocussed with respect to the aims of this study (e.g., “When I go to a party, I usually expect not to enjoy myself”). In contrast, the PIP items were specifically focussed on caring for a child with a health condition and so were deemed more appropriate. In addition, the PSI is comprised of 36 items, and it was considered that this may cause fatigue in participants. The PIP was selected for use in this study, to reduce response burden.

Psychological Flexibility

Initially two scales were considered for the measurement of psychological flexibility in parents. Both were informed by the Appearance and Action Questionnaire (AAQ-II, Bond et al., 2011), a widely used measure of psychological flexibility. The first was the Parenting-Specific Psychological Flexibility (PSPF) scale (Brassell et al., 2016), a 7-item scale created by adapting the item content of the original AAQ-II to reference the role of a parent. However, this scale focussed exclusively on the cognitive defusion aspect of psychological flexibility (see section 5.4.1. for further details), whereas parents in the first study of the PhD had reported other facets of psychological flexibility in their lives (e.g., committed action, see section 5.4.1. for further details).

The second scale considered was the Parent Psychological Flexibility (PPF) questionnaire (Burke & Moore, 2014), a 19-item scale with three subscales: cognitive defusion, committed action, and acceptance. Items are rated on a 7-point Likert scale from never true (0) to always true (6). This representation of different facets of psychological flexibility was judged a more accurate reflection of experiences reported by parents in study 1. Consequently, the PPF questionnaire seemed more appropriate to measure psychological flexibility. Each subscale had acceptable internal consistency in the present sample (cognitive defusion, $\alpha = 0.88$, committed action, $\alpha = 0.65$, acceptance, $\alpha = 0.76$) and the overall measure had good internal consistency ($\alpha = 0.88$). All subscales were positively and significantly correlated with the AAQ-II, suggesting the scale has construct validity. It has also been used in the wider paediatric literature with parents of children with chronic pain, with all subscales demonstrating good internal consistency (α > 0.87, Wallace, McCracken, Weiss, & Harbeck-Weber, 2015b).

Self-Compassion

The Self-Compassion Scale-Short Form (Raes et al., 2011) is a 12-item measure of self-compassion. The items are rated on a 5-point Likert scale ranging from almost never (1) to almost always (5) (Raes et al., 2011). The SCS-SF total score and the long form SCS total score have near perfect correlation (r = 0.98, Raes et al., 2011). Correlations between the long- and short-form subscales (on corresponding dimensions) were also very highly correlated. When used in the present sample the SCS-SF was found to have good internal consistency (α = 0.87).

The SCS-SF has previously demonstrated good internal consistency (α = 0.74) in a study of parental adjustment following a paediatric burn injury (Hawkins et al., 2019). A potential limitation is that it was initially developed using a convenience sample comprised of predominantly white undergraduate students (Raes et al., 2011). However, it has now been validated in a variety of settings (e.g., Meng et al., 2019), and used in a burn injury population. Therefore, the initial sample selected for development is unlikely to pose a threat to validity in the present study.

Social confidence

The CARe Burn scales are Patient Reported Outcome Measures (PROMs) designed to measure health and quality of life outcomes, specifically in individuals who have sustained a burn injury (Griffiths et al., 2019). There are four versions of the PROMs, including a scale for parents to complete about their experiences related to their burn injured child. The first study of this PhD found that parents of children with a range of visible differences have common experiences. One of these was anxiety about how to manage social interactions relating to their child’s visible difference, such as comments and questions from others. The CARe Burn Scale social situation subscale (3 items, rated on a 5-

point Likert scale) was thought to accurately reflect the concerns of these parents. Consequently, the CARe Burn scale social situation subscale from the parent version of the questionnaire was adapted for use with a cross-condition visible difference sample (e.g., changing the wording of items that refer to "burn injury" to instead refer "condition or injury"). In the present sample, this scale demonstrated good internal consistency (α = 0.85).

The CARe burn scales also correlate with other quality of life measures in burn injured individuals such as the Burn Specific Health Scale Abbreviated (Munster et al., 1987). This suggests it is a reliable and valid measure of various aspects of quality of life in a visible difference population. However, as this scale was developed specifically for parents and carers of burn injured children, it is yet to be validated with other visible difference samples.

Appearance investment

The Appearance Orientation subscale of the Multidimensional Body-Self Relations Questionnaire (MBSRQ) (Cash, 2000) was selected to measure the extent to which parents are invested in their own appearance. It is a 12 -item scale, each rated on a 5-point Likert scale rating from definitely disagree (1) to definitely agree (5). The Sociocultural Attitudes Towards Appearance Questionnaire-3 (SATAQ-3, Thompson et al., 2004) that measures internalisation of appearance ideals was also considered, but on close examination the items measuring appearance investment in the MBSRQ better reflected, the experiences reported by study I participants For example, parents reported self-evaluative feelings and appearance fixing behaviours, relating to both themselves and their child, as well as commenting on the role of appearance in self-worth. This is more indicative of investment in appearance rather than internalisation of appearance ideals.

The appearance orientation subscale demonstrated good internal consistency in the present sample (α = 0.89), and in both male and female participants in previous research in the general population (Males: α = 0.88, Female: α = 0.85, Cash, 2000). This subscale has demonstrated good test-retest reliability over a one-month period in both male and female participants (male: $r = 0.89$, female: $r = 0.91$, Cash, 2000). It has previously been used with a sample of individuals with scleroderma, a progressive connective tissue disease which causes significant appearance-related changes (Heinberg et al., 2007). The appearance orientation subscale has also been used in a number of studies exploring parental influence of body image (Galli et al., 2014; Schwartz et al., 1999), but has not been used directly with parents.

Perceived social support

The Multidimensional Scale of Perceived Social Support (MSPSS) is a 12-item measure of subjectively assessed social support (Zimet et al., 1988). Each item is answered on a 7-point Likert scale ranging from very strongly disagree (1) to very strongly agree (7). It has three subscales, each addressing a different source of social support: 1) Family, 2) Friends and 3) Significant Other. All subscales were found to have good internal consistency in previous research (family $\alpha = 0.87$, friends $\alpha = 0.85$ significant other = 0.91), and the full scale had good internal consistency in the present sample ($\alpha = 0.94$). The subscales also have good test-retest reliability over a 2-3 months period (family $\alpha = 0.85$, friends $\alpha = 0.75$, significant other $\alpha = 0.72$, Zimet et al., 1988). The MSPSS was also found to be negatively correlated with the depression and anxiety subscales of Hopkins Symptom Checklist (HSCL), suggesting that this scale does have construct validity (Zimet et al., 1988).

The Interpersonal Support Evaluation List (ISEL, Cohen, 1983) was also considered, but it contains 40 items and was deemed too burdensome. In addition, the items within the scale often referred to specific situations (e.g., “It would be difficult to find someone who would lend me their car for a few hours”) and so may not give a good overall representation of parents’ social support network.

The MSPSS is prevalent in research among the wider paediatric literature, including studies with parents of children with an enzyme disorder or intellectual disabilities (Grant et al., 2013), leukaemia (Nursyamsiyah, 2019), type 1 diabetes (Monaghan et al., 2011), and cerebral palsy (Wang et al., 2017). This scale only measures social support from three sources. When considering the literature on social support, social support not only has many sources, but also a number of functions (e.g., practical support, emotional support; Zimet et al., 1988). Consequently, there may be limitations to the different types of social support that this scale captured. To address this limitation, the qualitative data was examined to identify any alternative sources or functions of social support not captured by the MSPSS.

Optimism

The Life Orientation Test Revised (LOT-R**)** is a 10-item measure assessing generalised expectancies for positive versus negative outcomes (Scheier et al., 1994). This scale is used to provide a measure of an individual’s optimism. Items are answered on a 5-point Likert scale, ranging from strongly disagree (0) to strongly agree (4). LOT-R scores have been significantly correlated with different aspects of coping as measured by the COPE scale, - active coping, planning, positive interpretation and growth (Scheier et al., 1994) - suggesting it has good construct validity. The scale had good internal consistency in the present sample ($\alpha = 0.86$) and has previously been used in the visible difference research. For example, with 1,163 parents of children with CLP (Stock et al., 2020), where LOT-R scores were significantly correlated with outcome measures of parent wellbeing.

A criticism of the LOT-R is that by using it as a two-factor scale (optimism and pessimism), it conflicts with the original author’s theoretical definition of the scale (Scheier et al., 1994). Pessimism and optimism have been previously described as the polar opposites of a continuum. Therefore, in a recent review the original authors recommended that the LOT-R be used as a unidimensional scale (Carver et al., 2010). This advice was followed in the analysis of the present study.

Knowledge about condition and satisfaction with treatment

The Clinical Excellence Network-Questionnaire (CEN-Q) is a scale designed to measure parent perceived knowledge and understanding of their child's visible difference and the treatment they have received, as well as satisfaction with treatment (Stock et al., 2016). Five items were adapted from this scale to assess the role of knowledge of condition/injury and satisfaction with treatment. Items are answered on a 5-point Likert scale ranging from never (0) to almost always (4). The CEN-Q was developed specifically for use in a CLP population based on existing literature, clinical input and public involvement (Stock et al., 2016). Given that study 1 found that parents of children with a range of visible differences have common experiences, this measure was adapted by the PhD student for use in a cross-condition parent population. However, as it was developed specifically for CLP research, it is yet to be validated with other visible difference samples. The scale was found to have acceptable internal consistency in the present sample ($\alpha = 0.68$).

Study specific measures

As the population under investigation is a small and currently under- researched group, many existing measures do not accurately capture the experiences of these parents. Existing measures were altered, or study specific measures were created for the following constructs: perceived social challenges, perceived noticeability, parent self-efficacy, parent-child communication, and appearance fixing behaviours. All constructed items were reviewed by PPI members.

Perceived social challenges

Parents in study 1 discussed both actual and anticipated social challenges related to their child's appearance. Consequently, it was important to measure parent perceptions of the frequency and difficulty of the appearance-related social challenges that their child was currently experiencing. To capture this, items were adapted from project EAT III Teasing scale (Neumark-Sztainer et al., 2007), which has been widely used in body image and visible difference literature. One item was added to ask parents about the emotional impact of any teasing on themselves (“How upset were you about your child being teased?”). This was in addition to an existing item that was adapted to measure parents’ perceptions of how upset their child was about being teased. Items were answered on a 5-point Likert scale, ranging from never (0) to always (5). The total adapted scale was 3 items and was found to have good internal consistency in the present sample (α = 0.84).

Perceived noticeability

Parents in study 1 varied regarding how noticeable they reported their child’s visible difference to be. This was based on a variety of factors including perceived severity of the difference, how easy it was to conceal, how much they noticed it and how much other people commented on it. In the visible difference literature, subjective noticeability is typically related to appearance distress (Clarke, 2014). A single item measure of noticeability (e.g., “How noticeability is the condition to other people?”) has been used in previous visible difference literature (e.g., Zucchelli et al., 2020). This item was adapted for use in the present survey by changing the phrase “the condition” to “your child’s condition”. One further item was included to measure how noticeable the child’s condition or injury was to the parents themselves. These two items measuring different aspects of perceived noticeability were included in the analysis as two single item measures (“How noticeable is your child’s condition or injury to you?” and “How noticeable is your child’s condition or injury to other people?”). Items were scored on a 10-point Likert scale ranging from not at all noticeable (0) to very noticeable (10).

Self-efficacy

Existing measures of parent self-efficacy, such as the Tool to Measure Parenting Self-Efficacy (TOPSE, Kendall & Bloomfield, 2005), did not adequately reflect the experiences reported by parents of children with visible differences. Eleven items to measure self-efficacy were therefore constructed based on specific concerns and experiences described by parents in study 1. Some example items included: “I can support my child in telling others about their condition or injury", "I can promote confidence and resilience in my child" and "I can support my child in making decisions about treatment for their condition or injury". Focus on these specific concerns aimed to capture parenting self-efficacy related to their role as a parent of a child with a visible difference. Items were answered on a 5-point Likert scale ranging from not at all confident (0) to very confident (5). This scale had high internal consistency in the present sample (α = 0.90).

Parent-child Communication

Existing measures of parent-child communication, such as the Parent-Adolescent Communication Scale (PACS, Barnes & Olson, 1985), did not reflect the specificity of the experiences reported by parents of children with visible differences. Four items to measure parent–child communication were therefore constructed based on study 1 findings. These included: "I feel ok answering my child's questions about their difference in appearance" and "I feel ok talking to my child about treatment that will alter their appearance". Maintaining a narrow focus on parent-child communication related to the child's visible difference helped to ensure that parent responses were exclusively related to communication about their child's condition or injury. Items were answered on a 5-point Likert scale, ranging from never (0) to always (5). This scale had good internal consistency in the present sample (α = 0.87).

Appearance fixing behaviours

The Body Image Coping Strategies Inventory (BICSI; Cash, 2005) Appearance Fixing subscale has been used in previous visible difference literature and was considered for measuring appearance fixing behaviours in parents. However, scale items did not reflect the specific experiences reported by parents in study 1, such as the anticipatory anxiety related to specific places or situations in which their child's visible difference might be seen by others. In addition, adapted items asking a parent to reflect on the appearance of their child (e.g., "I make a special effort to make my child look their best"). was inappropriate given the context of the research and did not reflect parents' experiences reported in the qualitative data. A study-specific scale with four items was constructed to assess parent-reported appearance fixing behaviours. Items included "I attempt to cover or hide my child's difference in appearance" and "I feel uncomfortable in situations where my child's difference in appearance might be seen". Items were answered on a 4-point Likert scale ranging from definitely not like me (0) to definitely like me (4). This scale had good internal consistency in the present sample (α = 0.74).

Qualitative survey questions

Open-ended questions were included at the end of each topic section of the survey (e.g., Do you have anything else you would like to share related to communication about your child's condition or injury?").to give participants the opportunity to elaborate on their experiences, provide context or greater depth of their answers, and raise issues they felt were not captured via the existing questions (O'Cathain & Thomas, 2004).

Open-ended qualitative questions were included at the end of the survey to explore experiences of caring for a child with an appearance-affecting condition or injury during the COVID-19 outbreak and lockdown. (See Appendix E for reflections on the impact of COVID-19).

6.2.3. Piloting and public involvement input

The full survey was shared with the parent advisory group and three advisors completed it and provided feedback on content and design. Several changes were made following this feedback. For example, advisors suggested changes to the introduction to several measures to clarify how to respond to the questions. After these changes were made, the survey was piloted by an experienced researcher in the field who was external to the supervision team. This researcher was also the parent of young children so was able to reflect on the content from a professional and parenting perspective. Following this pilot, minor changes were made to the design (e.g., ensuring all the multiple-choice options fit on one screen).

6.2.4. Recruitment

Any parent of a child (aged 0-18 years old) with a condition or injury affecting their appearance and living in the UK was eligible to take part. Participants required a good understanding of written English to complete the survey. Due to variation in national healthcare systems, treatment pathways, and access to support (Wendt, 2009), parents based outside of the UK were excluded.

The initial plan was to recruit a broad sample, utilising several different online and face-to-face (e.g., charity conferences) recruitment strategies but due to the COVID-19 pandemic recruitment was restricted to online only (See Appendix D, for reflections on the impact of COVID-19). To address this potential limitation, and any possible sampling bias that may occur from targeting a single online platform. several different online platforms were utilised.

Various organisations were contacted, and 11 organisations agreed to advertise the study via their websites and e-newsletters. This included the member charities of the Appearance Collective, who support families of individuals affected by appearance-affecting conditions and injuries. The study was also advertised on the CAR social media pages (Twitter, Instagram, Facebook) and participant pool. To reach a more diverse parent population, who may not be connected to appearance collective charities, national and regional parenting support charities and organisations were also contacted and agreed to advertise the research (e.g., Mumbler sites, private regional parent support groups on social media). Organisations that provide specific support to single parents, foster parents, and adoptive parents were asked if they would advertise the study to their members. One charity who support single parents and one charity who support adoptive parents agreed to advertise the study. Finally, the online forum platform Reddit was used to post adverts on relevant forum pages (e.g., Cleft lip and palate forum, Eczema forum, Single parenting forum). Security measures were added into the survey design to prevent fraudulent behaviour and data were screened for inattentiveness (e.g., surveys completed in under 10 minutes).

6.2.5. Procedure

Parents and carers of children and young people with a visible difference were invited to take part in the online survey. Potential participants received information about the study via a brief advert or the recruitment video (Click here to see video). The use of short videos can support recruitment (O'Connor et al., 2014), and the use of recruitment videos can promote role modelling (Hendrickson, 2007). It was hoped that a recruitment video, including a testimonial from a parent who had completed the survey, might encourage others to take part. Parents were invited to click on a Qualtrics link which took them directly to the survey, where they were presented with a detailed information sheet.

Once the participants had read the information sheet, they were asked to read statements relating to the content of the information sheet and provide their consent to participate. Before beginning the main questionnaire, participants were also asked to complete several screening questions to confirm their eligibility. Screening questions asked parents if 1) they had a child with an appearance-affecting condition or injury, 2) if they were based in the UK, and 3) if they had a good understanding of written English. Those who did not meet the eligibility criteria were thanked for their time and informed that they unfortunately did not meet the participation criteria.

Eligible participants were asked to generate a unique code which would be used to anonymously identify their data, if they wished to withdraw from the research. Participants then completed the measures outlined above with an opportunity to provide more detail to their responses through open ended questions at the end of each section of the survey. At the end of the survey, participants were thanked for their time and invited into a prize draw to win a £50 Amazon voucher. They were also given the opportunity to indicate if they would like to receive a summary of the results.

Research ethics

An ethics application was submitted to the Faculty Research Ethics Committee and was returned with minor conditions to be addressed (e.g., including a telephone number as an alternative contact method). See section 3.8. for a discussion of general ethical considerations.

6.2.6. Participants

In total, 209 parents took part (*M* age = 36, SD = 7.26), of these 186 were female and 23 were male. Most were married or in a civil partnership (72%) and parenting within a traditional two parent family structure (89%). The majority of the sample was White British (71%). Parents provided demographic details about their child with a visible difference; 54% of the children with visible differences were female, 45% were male and the mean child age was 6 years old (SD = 4.93), with ages of children ranging from one month to 18 years old. See Table 7 for a summary of demographics.

Within the sample, n=19 (9%) parents also reported having a visible difference themselves. Of these parents, n=8 (42%) reported having a skin condition, n= 4 (21%) had a craniofacial condition, n= 4 (21%) had a birthmark, n=2 (11%) had scarring and n=1 (5%) reported having a condition that caused facial paralysis. This data provided helpful context as to what proportion of parents in the sample had had previous experience of living with a visible difference.

Table 7: Demographic information for survey participants (n=209)

	Mean	SD
Parent age	36	7.26
Child age	6	4.93
	N	%
Parent gender		
Female	186	89

Male	23	11
Child gender		
Female	113	54
Male	94	45
Prefer not to say	2	1
Relationship to child		
Mother	185	88
Father	23	11
Non-parent carer	1	< 1
Marital status		
Married/civil partnership	150	72
Cohabitating	37	18
Single	10	5
Divorced/separated	8	4
Did not report	3	1
Ethnicity		
Asian Other	1	<1
Black African	3	1
Mixed/multiple	5	2
South Asian	7	4
White British	149	71
White European	18	9
White Irish	9	4
White Other	16	8
Other	1	<1
Highest level of qualification		
GCSEs	15	7
A Levels/HND/BTEC	31	15
Bachelor's degree	108	52
Master's degree	36	17
Doctorate degree/PhD	9	4
Did not report	10	5
Child condition/injury		
Abdominal difference	1	<1
Birthmark	73	35
Burn injury	19	9
Craniofacial condition	75	36
Hair loss	9	4
Limb difference	11	5
Paralysis	4	2
Scarring	2	1
Skin condition	15	7

6.2.7. Data analysis

6.2.7.1. Calculating sample size

A power calculation was conducted to identify the number of participants required. A G*power calculation (Faul et al., 2007) indicated that a total sample size of 199 would be sufficient to detect medium to small effect size, with 95% power. Therefore, the present sample size of 209 resulted in adequately powered analyses. According to the sample size calculations for a Barons-Kenny's test (1986), based on the strength of mediation pathways, the sample of 209 was adequate to achieve 80% power (Fritz & McKinnon, 2007).

6.2.7.2. Data screening

Analyses were conducted using IBM SPSS Statistics (Version 26). The data were screened and cleaned following a detailed study specific plan (see Appendix F). Partial responses were removed from the data set. In total there were 232 completed responses. Firstly, data were screened to determine whether participants met the inclusion criteria for the study. Based on the information provided, participants who reported that their child had a condition that did not affect their appearance were removed from the data set (n=4). Any participants who were from outside the UK were also removed (n=19). The main analyses only included participants who reached the end of survey (n=209).

Within completed responses, there remained a small amount of missing data. Frequencies were conducted to determine the percentage of values that were missing (2.60%). Upon inspection of the patterns of missing data using a pattern analysis, no clear patterns of data missing across variables were found. However, the qualitative data provided by parents indicated that some felt that certain questions did not apply to them, as their child was too young (see section 6.3.3.), and as a result they had not completed these questions. As the items were likely to be intentionally missed out by participants, the missing data were determined to be at the individual level (Newman, 2014). Given the small percentage of missing data, the likely reason for non-responses, and the lack of relationship between variables with missing data, it was determined that pairwise deletion was an appropriate method for handling missing data (Newman, 2014).

6.2.7.3. Statistical assumptions

The data were examined to ensure they met the eight statistical assumptions required for a multiple regression (Field, et al., 2009). The assumption of linearity was tested using a scatter plot of the studentised residuals against the unstandardised predicted values, this assessed whether the collective independent variables of each regression model were linearly related to the dependent variable. Partial regression plots were then inspected to check for a linear relationship between the dependent variable and each independent variable in each regression model. All plots were found to indicate linear relationships. Data were also assessed for homoscedasticity by inspecting the scatterplots. Homoscedasticity was observed in these plots. Data were assessed for normality using Quantile-Quantile plots (Q-Q plots). Inspection of the Q-Q plots did not indicate deviations from model assumptions. Multicollinearity was assessed using Tolerance and VIF values. In all models, Tolerance values < 0.1 and VIF values were < 10, indicating no multicollinearity. Durbin-Watson statistics were run for each individual multiple regression model to test for independence of observations (reported below). Finally, the data were assessed for outliers and leverage points assessed by observing studentised deleted residuals, leverage points and Cook's distance measure of influence (reported below).

6.2.7.4. Main analyses

Multiple regression analysis

Multiple regression models were run to determine whether a number of predictor variables would significantly predict variance in an outcome variable. The outcome variables for the four regression models were: 1) parent negative affect, 2) parent positive affect, 3) stress frequency, and 4) stress difficulty. Purposeful regression modelling was selected in order to identify risk and protective factors in this population (Hosmer &

Lemeshow, 2000). Purposeful regression modelling is an augmented backwards elimination method in which the analyst makes systematic decisions about variable selection at each stage of the modelling process (Bursac et al., 2008).

The purposeful regression modelling protocol includes an initial stage of univariate analysis between the candidate predictor variables and outcome variable to determine whether predictors met the variable selection criteria to be include in the models. It has been found that the use of traditional p values (0.05 or 0.01) as a selection parameter in regression modelling can result in the exclusion of important variables (Bendel & Afifi, 1977; Mikey & Greenland, 1989). As such, these smaller p values are only recommended for very large sample sizes (events per variable of 100 or above), and values such as 0.20 or even 0.5 have been suggested as alternatives (Bursac et al., 2008; Heinze & Dunkler, 2017). Recommendations from the literature on selection parameters for regression modelling vary based on whether the candidate variables are based on previous knowledge (Heinze & Dunkler, 2017). Based on the relatively small sample size of the present research and the rigour used to select candidate variables, a selection parameter of $p < 0.20$ was selected for regression modelling in the present research. Any candidate variables that met this selection criteria during univariate analysis stage were entered into the models and then iteratively reduced using backwards elimination. Univariate regression analyses were carried out for all continuous potential predictor variables, whereas an independent samples t-test was utilised for the categorical variables: gender and type of condition (congenital or acquired).

Mediation analysis

To further explore the mechanisms underlying relationships identified in the regression analysis, two mediation analyses yielding 5000 bootstrap samples were conducted (Hayes, 2017). Hayes' PROCESS software was used to carry out this analysis.

The first mediation analysis was conducted to explore whether self-compassion was a mediating factor in the relationship between psychological flexibility and stress in parents of children with a visible difference. This mediation analysis was conducted because psychological flexibility was retained in the purposeful regression modelling process but did not have a significant relationship with stress difficulty in the final model, as hypothesised. The analysis was conducted to further explore and clarify this relationship. The theoretical overlap between psychological flexibility and self-compassion has been well documented, and there is empirical evidence to indicate associations between the two psychosocial constructs, and with psychological wellbeing (Marshall and Brockman, 2016). These two constructs were also observed to be correlated in the final regression model for stress difficulty. Furthermore, existing literature in the visible difference field (Hawkins et al., 2019; Shepherd et al., 2019; Zucchelli et al., 2020) and the present PhD findings have stated the importance of psychological flexibility and self-compassion for wellbeing in individuals and parents of children with a visible difference. Therefore, it was important to conduct this exploratory analysis to understand the possible role of psychological flexibility and self-compassion in supporting the wellbeing of parents.

Secondly, a mediation was conducted to explore the mechanism underlying the relationship between parent-reported teasing and positive affect. The mediation analysis was conducted because the relationship between teasing and positive affect was significantly positive rather negative, which was unexpected. The analysis was performed to investigate this relationship further. Existing research in the visible difference field has highlighted that parental confidence in their own ability to acquire skills and adapt in their parenting role are vital in supporting a child with a visible difference in a potentially challenging social situation (Horridge et al., 2010). Furthermore, the parent involvement model (Epstein & Van Voorhis, 2010) suggests that parent involvement can have an influence on a child's experience of school-based teasing (Cross et al., 2018). Teasing only had a significant relationship with positive parent affect when in the presence of other variables within the regression model. Teasing and social confidence were observed to be correlated in this model. Therefore, this exploratory analysis was conducted to investigate whether parental social confidence was a mediating factor in the relationship between teasing and positive affect in parents of children with a visible difference.

6.2.7.5. Content analysis

The qualitative data collected from open-ended questions in the online survey were analysed using deductive content analysis. Content analysis is a systematic and objective means of describing and quantifying data (Sandelowski, 1995). This method of analysis also allows researchers to test theoretical issues and build a conceptual model or system of a phenomena (Elo & Kyngas, 2008). This was an appropriate method for the present study because the qualitative data were used to enhance and clarify the understanding of constructs examined in the quantitative portion of the survey.

A deductive approach to content analysis is typically adopted when the aim is to test an existing theory or framework or to compare categories of data across different situations or time points, or otherwise examine data in a new context (Elo and Kyngas, 2008). The pre-existing categorical framework for this content analysis was informed by the psychosocial constructs under investigation in the online survey. Open-ended questions were included at the end of each section of the survey. Consistent with a mixed methods sequential approach (Creswell et al., 2007), each open-ended question related to a psychosocial construct identified within the findings of study 1 and existing theory and literature. Therefore, the qualitative data was coded using a categorisation matrix which reflected the predictor and outcome variables selected for the quantitative portion of this study. As described below (section 6.3.3.), “Impact of COVID-19” was a novel main category of the categorisation matrix, not drawn from the findings of study 1 or previous literature.

The first step of deductive content analysis is to develop a categorisation matrix (Elo and Kyngas, 2008). Either a structured or unconstrained matrix can be used, depending on the aim of the research. An unconstrained approach was selected for the present study, as this allowed new categories to be created within the existing matrix. Once the matrix was developed, the data were reviewed and coded within the identified categories (Polit and Beck, 2004). If data belonging to a novel category was identified, this data was coded using an inductive procedure: open coding (notes are written on the text whilst reading it), categories formed from open codes, and abstraction (formulating a description of the category and forming subcategories). To increase credibility of the analysis, the coding of qualitative data was discussed with members of the supervisory team and one external researcher (a postgraduate psychology student with experience of visible difference and body image research). The categorisation matrix was discussed with example quotes for each category and sub-category. If there was any disagreement coding or the conceptualisation of categories between researchers, this was discussed, a mutual decision was made, and these changes were integrated into the categorisation matrix. The researchers discussed and agreed on final coding, as well as conceptual code and category names. The categorisation matrix can be found in Appendix H.

6.2.7.6. Integrating quantitative and qualitative data

The aim of utilising a mixed methods design in the online survey was to collect qualitative data that would enhance and clarify aspects of the quantitative data. The qualitative and quantitative data were integrated using a method known as “following a thread” (Moran-Ellis et al., 2006). There is no standardised protocol for this method, but it involved identifying a theme or question in one data set and following it across to the other data set to enhance understanding. In this study, a theme or question was identified in the quantitative survey data and then followed into the corresponding qualitative open-ended questions to provide further detail. The quantitative and qualitative data will be discussed in tandem in greater detail in the discussion section of this chapter.

6.3. Results

6.3.1. Multiple Regression Models

Parent negative affect

Following preliminary analysis of the data, the following variables were included initially in the regression modelling process predicting *parent negative affect*: parent gender; child age; perceived noticeability to parents; appearance fixing behaviours; parent-reported teasing; parent-child communication; parent social confidence; self-compassion; optimism; psychological flexibility; social support; self-efficacy; and knowledge of condition and satisfaction with treatment. The backwards elimination process resulted in the removal of the following variables: parent gender, self-efficacy, psychological flexibility, social support. Following the addition of non-significant variables from the univariate analysis, perceived noticeability to others was retained in the model.

The model indicated independence of residuals, as demonstrated by a Durbin-Watson statistic of 2.21. There were no studentised deleted residuals greater than 3± standard deviations, no leverage points above 0.2 and all Cook's distance values were below 1 (Cook and Weisburg, 1982). The fitted model accounted for 33% of the variance and comprised of five statistically significant variables ($F(10,158) = 9.31$, $p < .05$, adj. $R^2 = .331$). Increased parent-reported teasing was positively associated with negative affect scores, whereas increased child age, greater parent-child communication scores, greater appearance investment scores, and greater knowledge of condition and satisfaction with treatment were all negatively associated with negative affect scores. See Table 8 for final regression model.

Table 8: Final multiple regression model for parent negative affect,

Predictor variable	B	Standardised coefficients β	Sig .	F	*df*	Sig.	Adj. R^2
Overall model				9.314	10,158	.000	.331
Child age	-.203	-.141	.046*				
Noticeability to parent	.279	.129	.061				
Noticeability to others	-.224	-.089	.192				
Appearance fixing	1.449	.138	.069				
Teasing	.946	.156	.033*				
Parent-child communication	-1.266	-.173	.014*				
Self-compassion	-1.497	-.155	.071				
Optimism	-.232	-.165	.052				
Knowledge and satisfaction with treatment	-1.800	-.175	.014*				
Appearance investment	-1.367	-.146	.024*				

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*= p < .05, ** = p < .01

Parent stress frequency

Following preliminary analysis, the following variables were included initially in the regression model predicting *parenting stress frequency:* perceived noticeability to parent; perceived noticeability to others; appearance fixing behaviours; parent-reported teasing; parent-child communication; parent social confidence; self-compassion; optimism; psychological flexibility; social support; self-efficacy; and knowledge of condition and satisfaction with treatment. The backwards elimination process resulted in the removal of the following variables: self-efficacy, parent social confidence, perceived noticeability to others, psychological flexibility, appearance fixing behaviours. See Table 9 for final regression model.

The model indicated independence of residuals, as demonstrated by a Durbin-Watson statistic of 1.92. There was one studentised deleted residual greater than 3± standard deviations. This response was retained as it was not deemed to be a data entry error, and there were no leverage values greater than 0.2 and all Cook's distance values were below 1. The fitted model accounted for 32% of the variance and was comprised of four statistically significant variables ($F(8,160) = 11.23$, $p < .05$, adj. $R^2 = .328$). Greater perceived noticeability to parents and increased parent-reported teasing were positively associated with stress frequency whereas, greater self-compassion and greater knowledge of condition and satisfaction with treatment were negatively associated with stress frequency in parents.

Table 9: Final multiple regression model for stress frequency

Predictor variable	B	Standardised coefficients β	Sig .	F	*df*	Sig.	Adj. R^2
Overall model				11.32	8,160	.000	.328
Noticeability to parents	.213	.225	.001**				
Teasing	.861	.325	.000**				
Parent-child communication	.331	-.103	.122.				
Self-compassion	.759	-.179	.036*				
Optimism	.078	-.127	.134				
Social support	.255	.100	.159				
Knowledge and satisfaction with treatment	.933	-.207	.002**				
Parent gender	.891	.631	.160				

*= p < .05, ** = p < .01

Parent stress difficulty

Following preliminary analysis, the following variables were included initially in the regression model for *parent stress difficulty*: child gender; perceived noticeability to parents; appearance fixing behaviours; parent-reported teasing; parent-child communication; parent social confidence; self-compassion; optimism; psychological flexibility; self-efficacy; and knowledge of condition and satisfaction with treatment. The backwards elimination process resulted in the removal of the following variables: self-efficacy, parent social confidence, child gender, optimism. See Table 10 for final regression model.

The model indicated independence of residuals, as demonstrated by a Durbin-Watson statistic of 2.00. There were no studentised deleted residuals greater than 3± standard deviations, no leverage points above 0.2 and all Cook’s distance values were below 1. The fitted model accounted for 29% of the variance and was comprised of four statistically significant variables (*F*(8,160) = 7.77, *p* < .05, adj. R^2 = .294). Increased appearance fixing behaviours and increased parent-reported teasing were positively associated with stress difficulty, whereas greater parent-child communication scores and greater self-compassion were negatively associated with stress difficulty.

Table 10: Final multiple regression model for stress frequency

Predictor variable	B	Standardised coefficients β	Sig.	F	*df*	Sig.	Adj. R^2
Overall model				7.77	8,160	.000	.294
Noticeability to parent	.152	.140	.051				
Appearance fixing	.944	.180	.027*				
Teasing	.734	.242	.001**				
Parent-child communication	-.624	-.170	.025*				
Self-compassion	-1.421	-.293	.001**				
Psychological flexibility	.689	.147	.096				
Knowledge and satisfaction with treatment	-.671	-.130	.00				
Social support	.300	.102	.172				

*= p < .05, ** = p < .01

Parent positive affect

Following preliminary analysis, the following variables were included initially in the regression model predicting *parent positive affect*: child age; child gender; appearance fixing behaviours; parent-child communication; parent social confidence; self-compassion; psychological flexibility; social support; self-efficacy; and knowledge of condition and satisfaction with treatment. Following the addition of non-significant variables from the univariate analysis, parent-reported teasing was retained in the model. See Table 11 for the final regression model.

The model indicated independence of residuals, as demonstrated by a Durbin-Watson statistic of 2.01. There were no studentised deleted residuals greater than 3± standard deviations, no leverage points above 0.2 and all Cook's distance values were below 1. The fitted model account for 23% of the variance and was comprised of four statistically significant variables ($F(5,167) = 11.32$, $p < .05$, adj. $R^2 = .231$). Increased child age was negatively associated with positive affect, whereas greater parent social confidence, greater parenting self-efficacy, and increased parent-reported teasing were positively associated with positive affect.

Table 11: Final multiple regression model for positive affect

Predictor variable	B	Standardised coefficients β	Sig.	*F*	*df*	Sig.	Adj. R^2
Overall model				11.32	5,167	.000	.231
Child age	-.598	-.295	.000**				
Child gender	1.943	.099	.148				
Social confidence	1.658	.170	.030*				
Self-efficacy	5.309	.331	.000**				
Teasing	1.79	.208	.006**				

*= p < .05, ** = p < .01

6.3.2. Mediation analysis

A mediation analysis was conducted to examine whether self-compassion mediated the relationship between psychological flexibility and stress difficulty. Figure 9 shows the path diagram for the mediation model tested. It was hypothesised that 1) psychological flexibility would be positively associated with self-compassion, 2) self-compassion would be negatively associated with stress difficulty and 3) self-compassion would mediate the relationship between psychological flexibility and stress difficulty.

Coefficients for each regression pathway, including direct effects between psychological flexibility and stress difficulty, are given. With stress difficulty as the outcome variable, psychological flexibility showed a significant indirect effect through self-compassion, $a*b$ = -.6728, 95% CI [-1.1067, -.2506]. In the presence of the indirect effect (ab = -.6728) of psychological flexibility on stress difficulty through self-compassion, the direct effect (c' = -.1961) was not statistically significant. The analysis suggests that self-compassion completely mediated the relationship between psychological flexibility and parenting stress difficulty.

Simple linear relationship

Mediated relationship

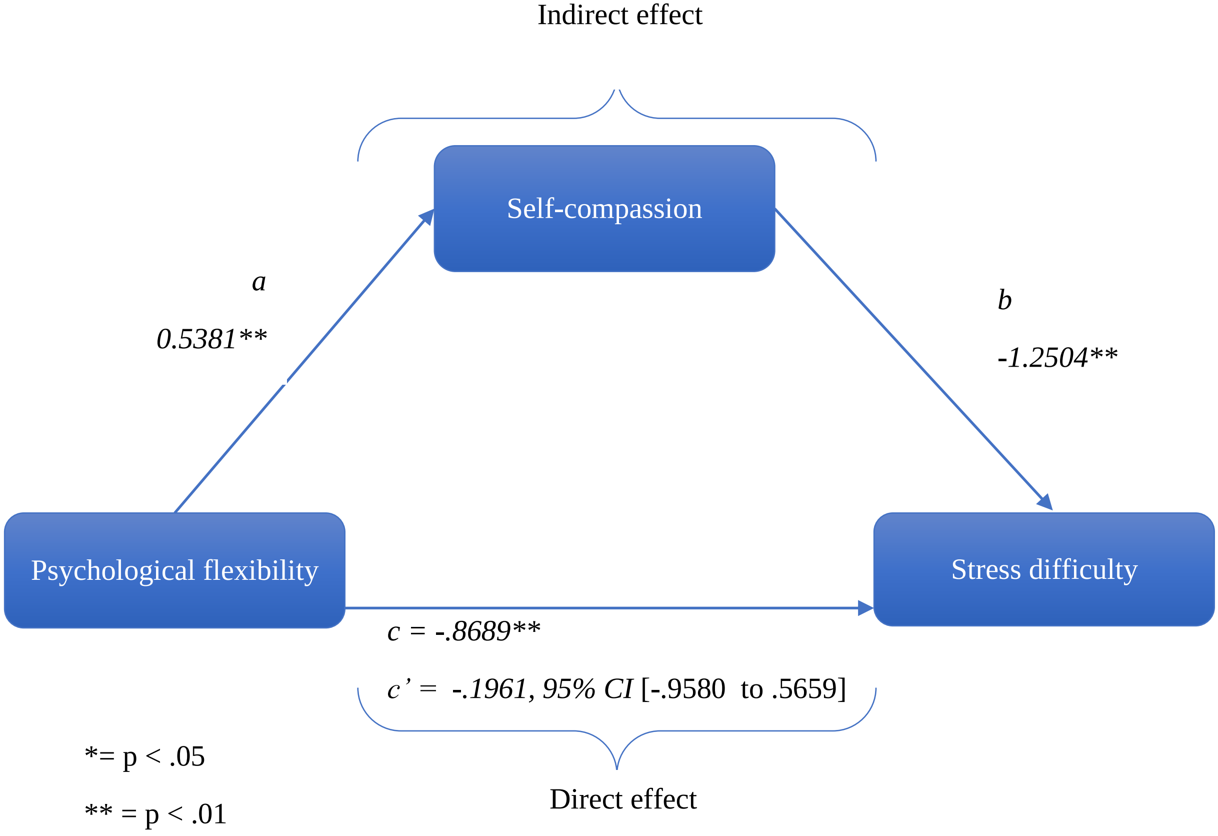


Figure 9: Diagram of simple and mediated model between psychological flexibility and stress difficulty.

a = effect of predictor on the mediator

b= effect of the mediator on the outcome

c = total effect of predictor on outcome without the mediator, total effect= direct + indirect effect

c' = direct effect of predictor on the model, controlling for mediator

ab = indirect effect of predictor on outcome through mediator

A second mediation analysis was conducted to examine whether parent social confidence mediated the relationship between parent-reported teasing and positive affect. Figure 10 shows the path diagram for the mediation model tested. It was hypothesised that 1) parent-reported teasing would be negatively associated with parental social confidence, 2) parental social confidence would be positively associated with positive affect and 3) parental social confidence would mediate the relationship between parent-reported teasing and positive affect.

Coefficients for each regression pathway, including direct effects between teasing and positive affect, are given. With positive affect as the outcome variable, teasing showed a significant indirect effect through social confidence, $a*b$ = -.3749, CI [-.8462, -.0478]. In the presence of the indirect effect (ab = -.3749) of teasing on positive affect through social confidence, the direct effect (c' = .1.0286) is not statistically significant. As the c' pathway is the opposite sign to *ab* pathway this is referred to as inconsistent mediation (MacKinnon et al., 2007). In an inconsistent mediation it is possible that step one of the mediation (pathway c) is not significant, but there is still mediation (MacKinnon et al., 2007). This is the effect demonstrated in the present analysis.

Simple linear relationship

Mediated relationship

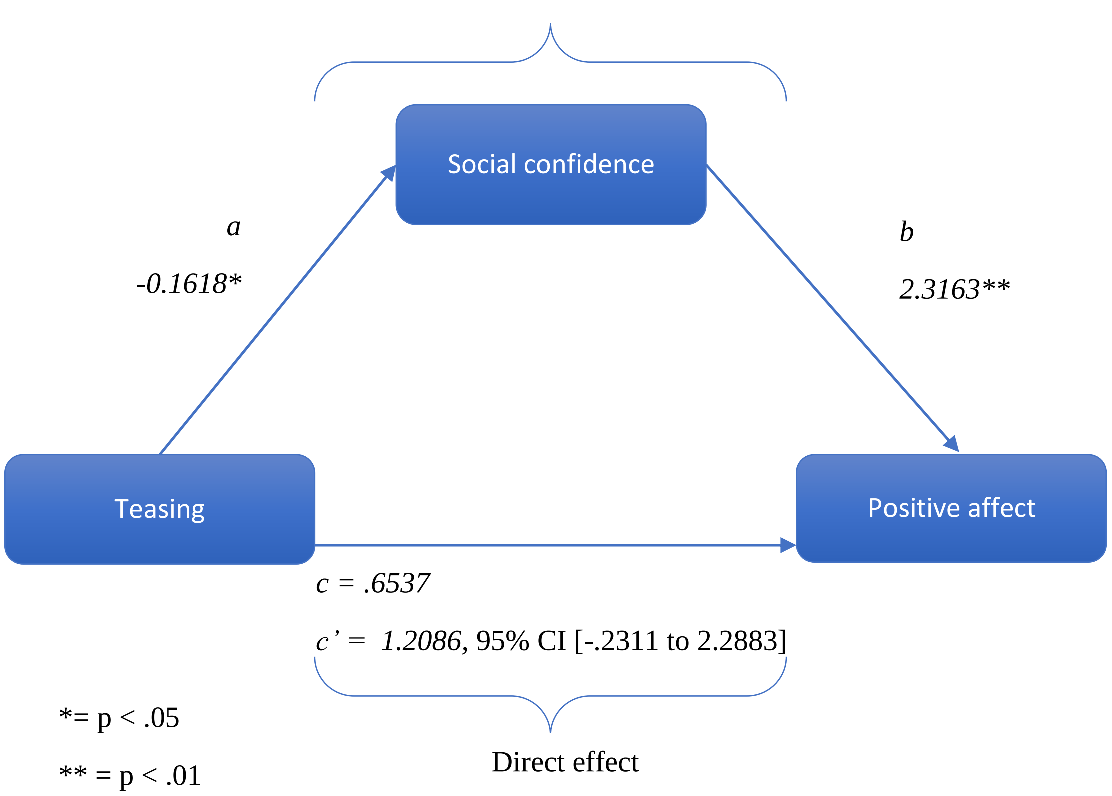


Figure 10: Diagram of simple and mediated model between teasing and positive affect

a = effect of predictor on the mediator

b= effect of the mediator on the outcome

c = total effect of predictor on outcome without the mediator, total effect= direct + indirect effect

c' = direct effect of predictor on the model while controlling for the mediator

ab = indirect effect of predictor on outcome through mediator

6.3.3. Content analysis

In total, 167 participants (80% of total sample) responded to at least one open-ended question within the survey. Seven main categories were identified, each with sub-categories (Elo and Kyngas, 2008). Six a priori categories were included in the original categorisation matrix: Parent affect and stress, Reactions of other people to child’s visible difference, Communication, Strategies for coping, Self-efficacy in parenting role, Appearance investment, Impact of COVID-19. Several novel sub-categories were identified inductively within the qualitative data and integrated (e.g., pre-existing mental health diagnoses within the stress/affect main category). The findings presented below supported the qualitative findings of study 1 and enhanced the quantitative data collected for the present study. The seventh category, “Impact of COVID-19”, was a novel main category not a topic covered in the previous study, which was conducted in 2019, prior to the beginning of the pandemic. This was included at this stage of the research to acknowledge and provide opportunity for parents to report on the challenges experienced during the COVID-19 outbreak and lockdown (see appendix E for further reflections on COVID-19).

Categorisation matrices were broken down into three levels: main categories, generic categories, and sub-categories. Although this has resulted in some sub-categories being appearing with low frequencies, this differentiation and close examination of the qualitative data provided a highly detailed insight into the varied experience of parents. All categories are described in detail below. A “following a thread” approach was used to integrate the findings from the quantitative and qualitative data sets. A description of this method can be found in section 6.2.7.6. See Figures 11-17 for a visual representation of the categorisation matrix and Tables 13-19 for a breakdown of the frequencies for each category. When describing the findings, guidelines around quantifying language were adhered to (Hill et al., 2005). “All” refers to all participants, “most” refers to more than half, “some” refers to less than half but more than two.

Parent affect and stress

Parents elaborated on their experiences of positive and negative affect, as well as other difficult thoughts and feelings associated with being a parent of a child with a visible difference. Parents reported low mood associated with feeling helpless and isolated in their experiences (n=17). Some also mentioned feelings of guilt (n=20), often related to the cause of their child's visible difference (e.g., an accident that caused a burn injury). Some parents provided detail about their experiences of anxiety (n=55) related to different aspects of their child's condition and injury, including concerns about the future (n=28), as well as generalised anxiety about their child's condition or injury (n=24). A few parents also reflected on the impact of their parenting role on their own pre-existing mental health difficulties (n=3). Stress-inducing aspects of caring for a child with a visible difference were also reported (n=8). This included the overall burden of care (n=5) and specific stressful experiences with healthcare services and professionals (n=3). In contrast, some parents reported the positive affect (n=14) that they associated with having a child with a visible difference (e.g., feelings of pride).

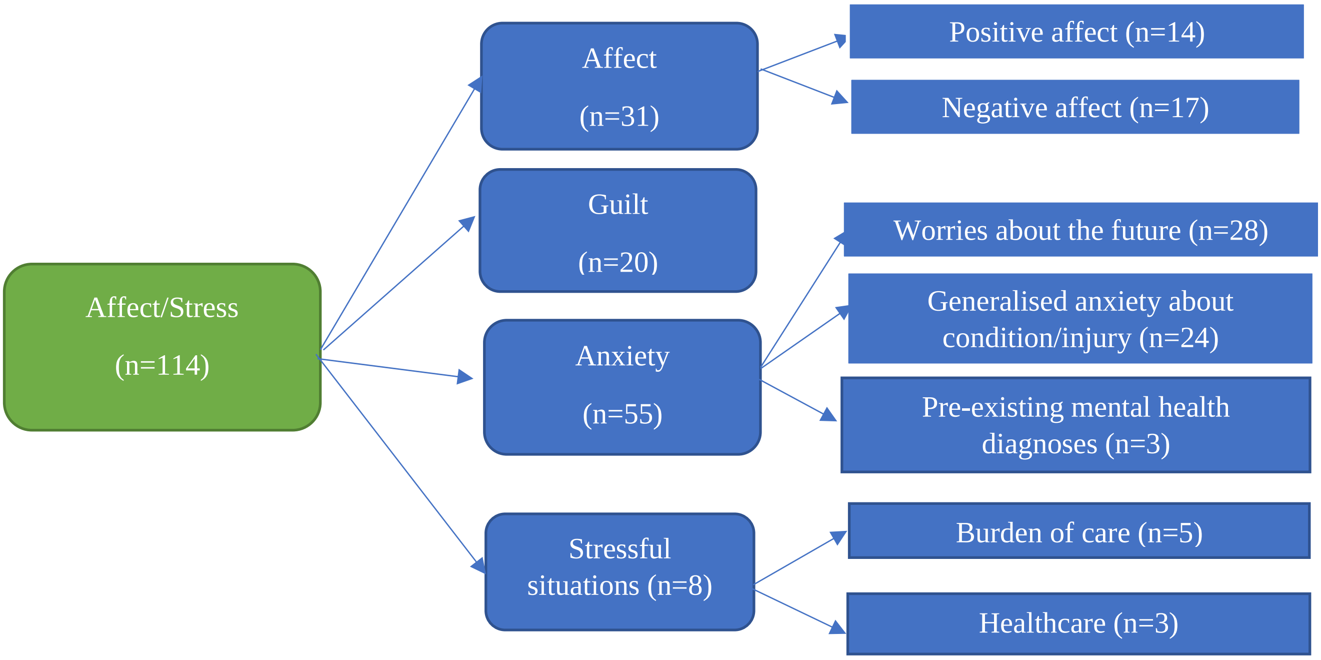


Figure 11: Categorisation matrix for affect/stress content analysis category.

Reactions of other people to child's visible difference

Parents reported the negative emotional impact of both actual interactions and anticipated social challenges on themselves (n=41), their affected child (n=3), and siblings (n=15). Most referred to a lack of awareness or understanding of their child's condition or injury (n=93), and consequently an emphasis on them to educate the public on behalf of their child. Some parents found taking on the role of educator empowering (n=29), whereas others struggled with questions, comments, and unsolicited advice from others (n=37). Some parents commented on the visibility of their child's appearance difference and how this is associated with the frequency, nature, and extent of other people's reactions (n=26). Some parents also reflected on the fact that they found that adults and older children tended to be less accepting of their child (n=27). Some also felt that they had little to add or report as their children were very young and had not yet been exposed to many social situations (n=26).

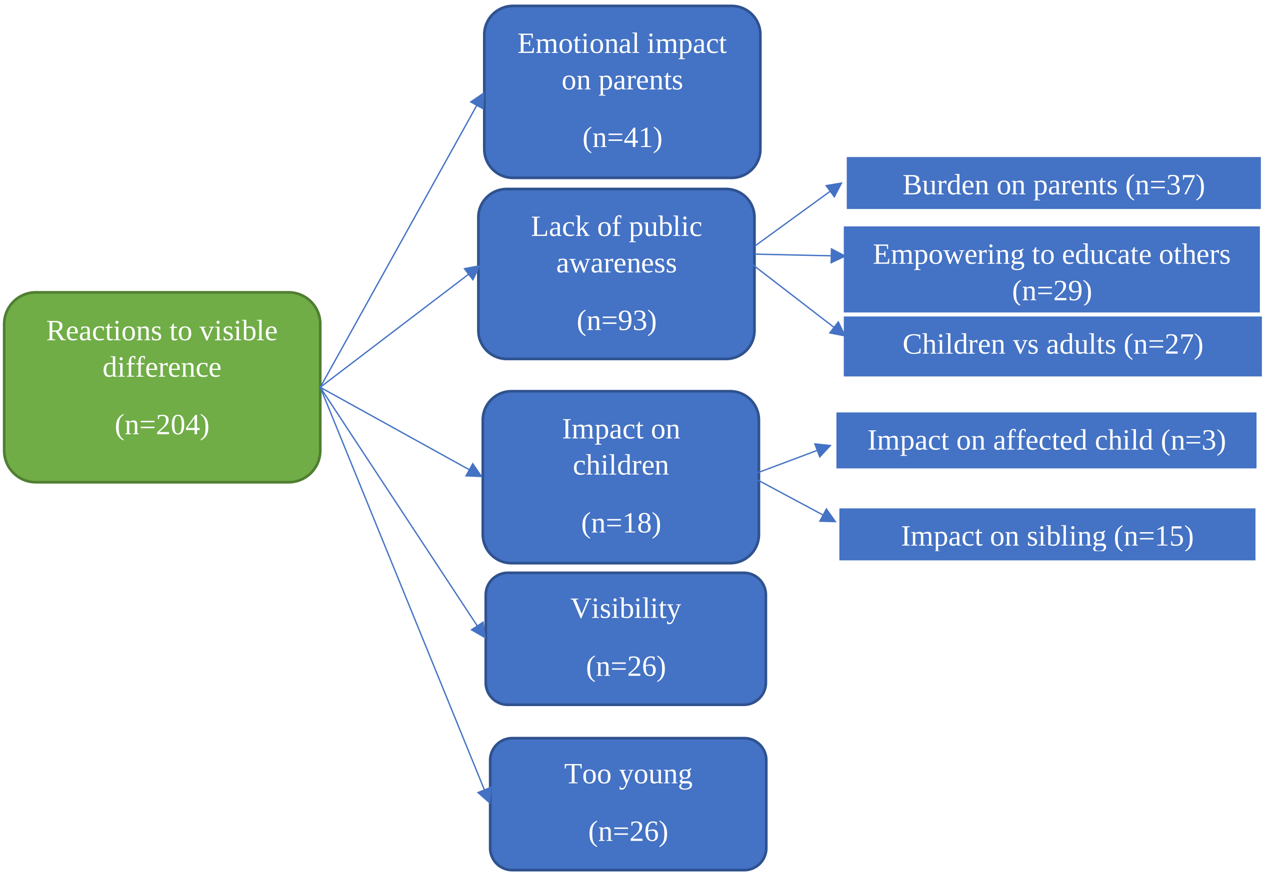


Figure 12: Categorisation matrix for reactions to visible difference category.

Communication

Some parents reported the importance of preparing their child to independently manage their condition and injury (n=38). This included having conversations about difference and diversity broadly (n=10), as well as educating their child about their specific condition or injury (n=15). They wanted to model how to communicate with others about their visible difference (n=13; e.g., handling questions and comments from strangers), and instil acceptance of the appearance difference (n=15), encouraging their children to embrace themselves as a unique individual. Some reported that their child was too young to engage in this form of communication (n=20).

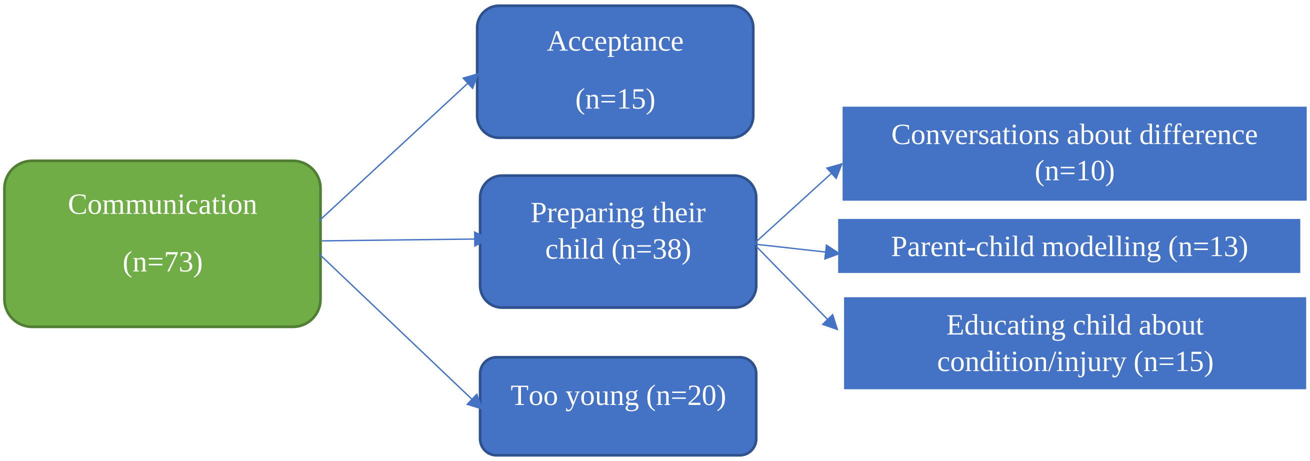


Figure 13: Categorisation matric for communication category.

Strategies for coping

Parents provided detail on the various strategies, techniques, and resources they draw on when coping with difficult thoughts and feelings related to caring for their child. Firstly, some parents spoke about active coping strategies (n=13), which involved actively seeking, reaching out or engaging in behaviour they knew would help them to cope. This included seeking help to support their own mental health by accessing therapy or other mental health services, reaching out to healthcare services/professionals and online platforms for information. In addition, a few parents reported that using their experiences to help others, also helped them to find purpose and feel connected to others (n=3). A couple of parents also expressed the importance of taking regular exercise to manage parenting stress (n=2). Some parents reported the psychological resources they felt had helped them to cope with difficult thoughts and feelings related to their parenting role (n=23). This included having an optimistic outlook (n=8), practicing self-compassion (n=4), and being resilient (n=11) when faced with challenges in their parenting role.

Finally, parents provided details on the role of social support (n=24) in helping them to cope with their difficulties. Support from friends and family (n=18) and peers (n=4) was invaluable. Whilst some also mentioned the important role played by charities and organisations in their ability to access social support (n=4), other parents felt that the support

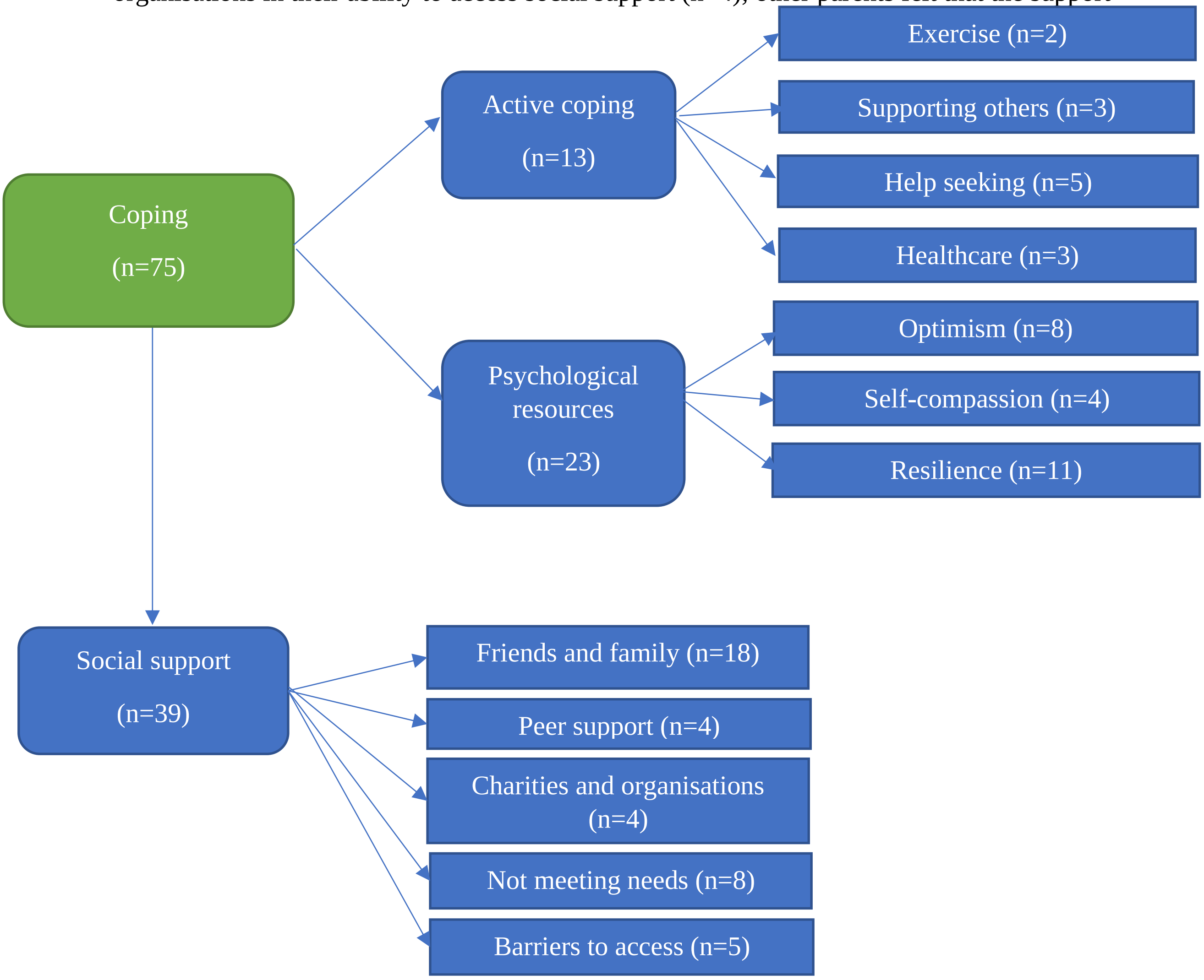


Figure 14: Categorisation matrix for coping category.

offered did not meet their needs (n=8) and barriers to accessing social support (n=5) were mentioned, including geographical distance, lack of information, and challenges sharing difficult thoughts and feelings with others.

Self-efficacy in parenting role

Parents provided detail on how equipped they felt to support their child and prepare them for managing challenges related to their child's visible difference. Some queried how best to support their child (n = 13). Of these, some parents who were taking supportive action remained concerned about whether they were "getting it right" (n = 5). Others reported that they needed more support to understand how best to support their child and how to implement this support (n = 8). Others reflected on the weight of responsibility they felt for getting this support "right", and the impact this realisation had on them (n=9). Some parents reported that they were confident in their knowledge and ability to support their child to manage their condition independently (n = 22).

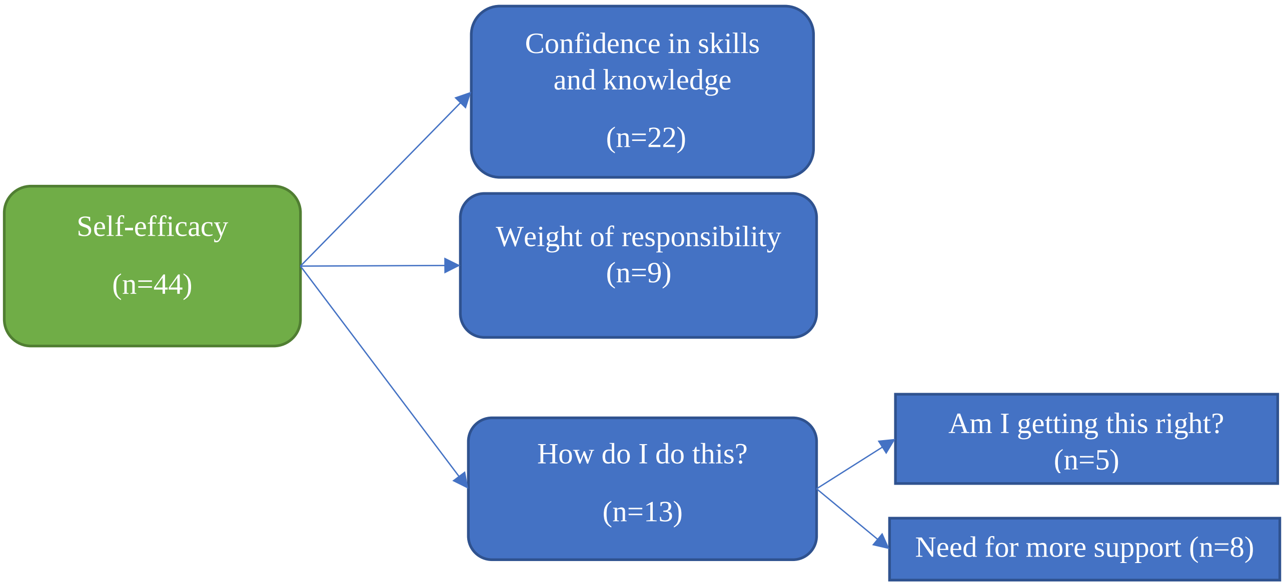


Figure 15: Categorisation matrix for self-efficacy category.

Appearance investment

Parents discussed their positionality on the importance of appearance as a personal attribute. Some reported that it was important to them (n = 17), for example liking to "take care" of themselves (n = 3) and 'look good' (n = 7). Two respondents described using appearance fixing behaviour (e.g., make-up) as a coping strategy when they felt upset or stressed, and two referred to the impact of narrow appearance ideals and how this shaped their thoughts and feelings about appearance (n = 2). Some mentioned their own difficulties with body image (n = 3), and several wanted to model positive body image to their children (n=9). In contrast, some reported that appearance was not important to them, and they did not prioritise it as something to spend time and energy on (n = 14).

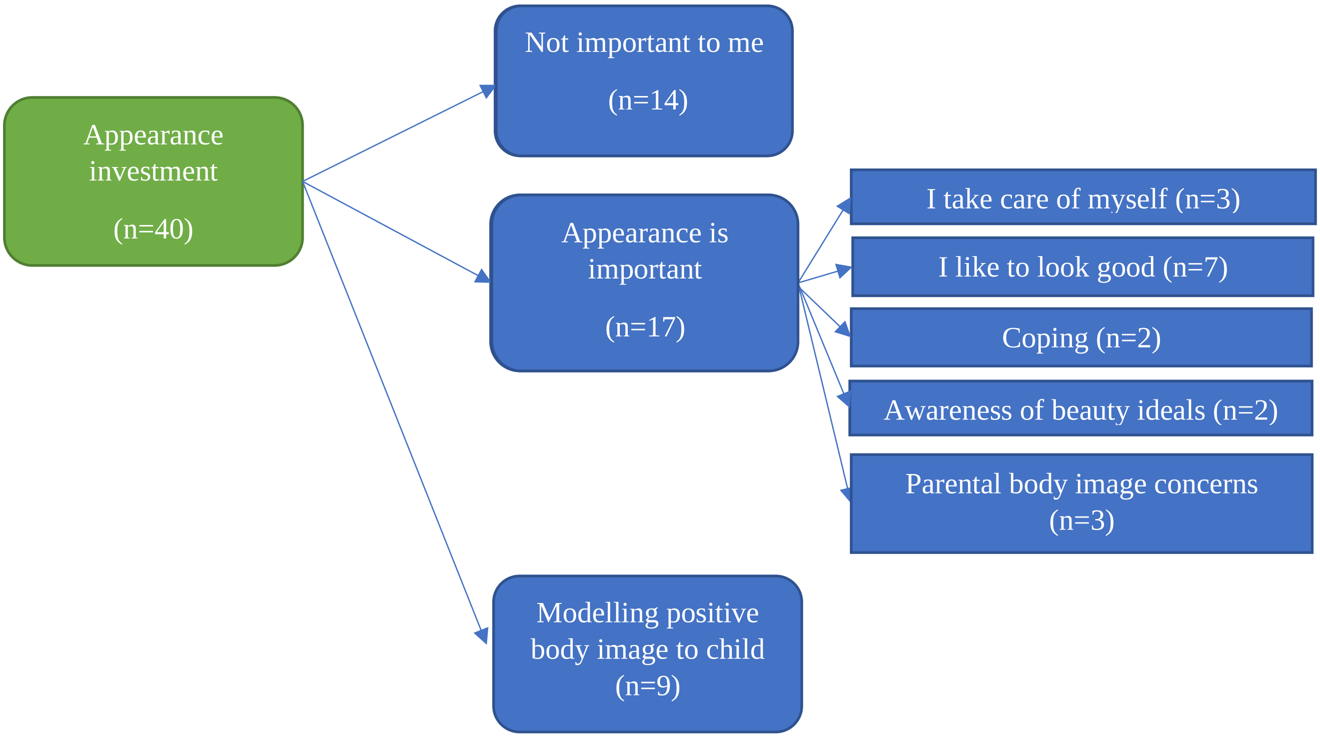


Figure 16: Categorisation matrix for appearance investment category.

Impact of COVID-19

Parents felt their access to support had been reduced because of COVID-19 (n = 11). This included support from both family and friends as a result of reduced social contact (n = 6), as well as fewer opportunities for formal support from health and support services due to reduced in-person support (n = 5). These data were collected towards the end of the first lockdown in the UK. Consequently, some organisations may not have had the opportunity to adapt their services to offer alternative, online support. Parents also had other concerns related to reduced social contact because of COVID-19 and lockdown (n = 50). Some were worried about their children becoming socially isolated (n = 11) and the potential impact of this on social skills development (n = 12). Some reflected on the potential impact of mask wearing (n = 8), and how this may be positive (e.g., less staring, fewer questions), whilst also feeling anxious about having to remove the mask. Many mentioned anxieties about the health risks of COVID-19 (n = 45), as some children also had underlying health conditions (n = 17). This anxiety was also sometimes exacerbated due to reduced access to healthcare during this time (n = 28). Finally, parents also reported the positives that had come from the COVID-19 lockdown for their families (n = 20), such as the ability to spend more quality time together.

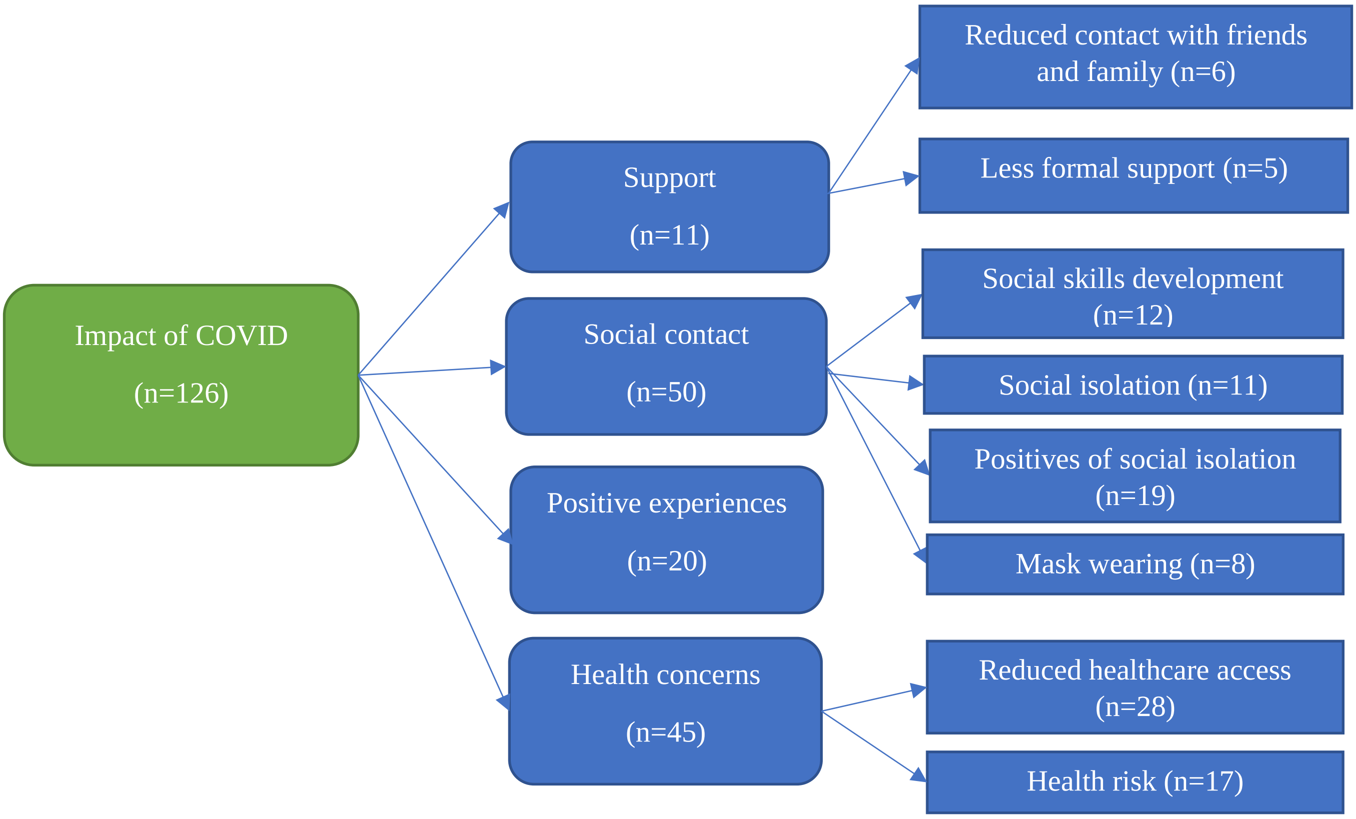


Figure 17: Categorisation matrix for impact of COVID category.

Table 12: Summary of frequencies for the parent affect and stress category

Category/Subcategory	Frequency *n*(%)
Parent affect and stress	67
Affect	31
Positive affect	14
Negative affect	17
Anxiety	55
Worries about the future	28
Generalised anxiety about condition/injury	24
Pre-existing mental health condition	3
Stressful situations	8
Burden of care	5
Healthcare	3
Guilt	20

Table 13: Summary of frequencies for the reactions to visible difference category

Category/Subcategory name	Frequency *n*(%)
Reactions to visible difference	131
Lack of public awareness	93
Emphasis on parents	37
Empowering to educate others	29
Children vs adults	27
Impact on children	18
Impact on affect child	3
Impact on sibling	15
Emotional impact on parents	41
Visibility	26
Child too young	26

Table 14: Summary of frequencies of the communication category.

Category/Subcategory name	Frequency *n*(%)
Communication	131
Preparing their child	38
Conversations about appearance	10
Parent-child modelling	13
Educating child about condition/injury	15
Promoting Acceptance	15
Child too young	26

Table 15: Summary of the frequencies for the coping category

Category/Subcategory name	Frequency *n*(%)
Coping	61
Active coping	13
Exercise	2
Supporting others	3
Help seekers	5
Healthcare	3
Psychological resources	23
Optimism	8
Self-compassion	4
Resilience	11
Social support	24
Friends and family	18
Peer support	3
Charities and organisations	4
Not meeting needs	8
Barriers to access	5

Table 17: Summary of frequencies for appearance investment category

Category/Subcategory name	Frequency *n*(%)
Appearance investment	33
Appearance is important	17
I take care of myself	3
I like to look good	7
Coping	2
Awareness of body ideals	2
Parental body image concerns	3
Not important to me	14
Modelling positive body image	9

Table 16: Summary of frequencies for the self-efficacy category.

Category/Subcategory name	Frequency *n*(%)
Self-efficacy	44
How do I do this?	13
Am I getting this right?	5
Need for more support	8
Weight of responsibility	9
Confidence in skills and knowledge	22

Table 18: Summary of frequency of the impact of COVID-19 category

Category/Subcategory	Frequency *n*(%)
Impact of COVID-19	126
Support	11
Reduced support from friends and family	6
Less formal support	5
Social contact	50
Social skills development	12
Social isolation	11
Positives of isolation	19
Mask wearing	8
Health concerns	45
Reduce health care access	28
Health risk	17
Positive experiences	20

6.4. Discussion

To increase the generalisability of the qualitative findings of study 1, this study aimed to assess the experiences of parents of children and young people with a visible difference using a cross-sectional survey design with measures intended to capture the experiences of psychosocial constructs identified in the qualitative data as important to the experience and coping strategies of parents. This study aimed to investigate possible risk and protective factors for parent affect and parent stress. Psychosocial constructs that had been identified as important to coping in the first study were used as predictors of variance in parent affect and stress outcomes. Finally, this study aimed to identify possible targets for intervention by gaining a better understanding of the risk and protective factors for parent distress. The main findings were that parent-reported teasing, noticeability to parents, and appearance-fixing behaviours were significantly positively associated with increased negative affect and stress. Whereas, self-compassion, good parent-child communication, knowledge of condition and satisfaction with treatment were significantly negatively associated with increased negative affect and stress. Self-efficacy and social confidence were found to be significantly positively associated with increased positive affect. The qualitative data provided further clarification and context for these findings. Parent qualitative responses most frequently reported on the following topics: other people's reactions to their child's condition, parent-child communication, and the impact of COVID-19. These findings will now be described below in greater detail, with reference to the broader literature.

6.4.1. Cross-condition findings

Based on Belsky's (1984) model of parenting, parenting psychological resources and contextual stress can indicate parental adjustment, as well as child wellbeing. The qualitative data indicated that 114 participants responded to questions about affect and stress, and 100 (88%) of these participants spoke about negative affect or stress. As a result, 48% of the overall sample provided qualitative data about their experience of negative affect and stress, in addition to their responses to the quantitative survey. Although some participants may have chosen not to respond to the open-ended questions, this suggests that there is variation in parent's experience of affect and stress.

As hypothesised, the present study found no significant difference in parent positive and negative affect and parenting stress between congenital and acquired visible differences. This supports the qualitative findings of study 1, that there are common cross-condition experiences across different appearance-affecting conditions and injuries. This also provides some evidence to support the overall aim of the PhD, to develop cross-condition support to meet identified unmet needs, based on risk and protective psychosocial factors identified in the data.

However, the multiple regression analysis found that these psychosocial factors only predicted between 23 and 33% of the variance in the parent affect and stress outcomes, The qualitative data indicated that there may be several factors which could contribute to this variance, which were not measured in the quantitative portion of the data collection. For example, some parents spoke about the impact of pre-existing mental health conditions on their ability to cope with challenges related to their child's condition or injury. Many parents also referred to the impact of COVID on their coping resources. Neither of these variables were included in the quantitative measurements as they had not been present in the themes of study 1. The impact of both pre-existing mental health conditions and stressful life events on parents is supported by empirical evidence within the visible difference field (Stock et al., 2020). Consequently, additional mental health challenges and stress, unrelated to their child's condition may also impact parent adjustment. This further highlights the complexities of parenting (Belsky, 1984), and suggests a need to consider the parent experience as a holistic whole.

6.4.2. Social challenges: actual and anticipated

An important finding of this study was that, as hypothesised, parent-reported teasing of their child by others was significantly positively associated with three of the four affect and stress outcomes (parent negative affect, stress difficulty and stress frequency) within the regression models. This indicates that parent perceptions of frequency and emotional impact of teasing is associated with parent negative affect and stress related to their parenting role. In addition, the qualitative data indicated that parents struggled with managing the reactions of other people and the emotional burden of coping with stigma, genuine misunderstandings, or ignorance, due to a lack of awareness and education of their child's condition among members of the public. This supports the findings of the first study of the PhD, particularly the sub-theme of "identify the threats", which described parents concern and awareness of possible social challenges that could be a threat to their child's wellbeing.

Appearance-related teasing is common in all children, both with and without a visible difference (Feragen & Stock, 2016). Negative reactions from others are commonly reported by individuals with visible differences (e.g., Hlongwa & Rispel, 2018; Albett & Thompson, 2016). Parents and carers of children with visible differences regularly report incidences or concerns about social stigma and exclusion (Albett and Thompson, 2016; Hlongwa and Rispel, 2018). A meta-ethnography of 12 studies to explore the psychosocial impact of skin conditions on both parents and young people (Albett and Thompson, 2016) found that young people identified themes of feeling different and some children expressed that teasing was the worst part of living with their condition. In addition, parents also reported fears of appearance-related teasing. The study also identified a broader theme, which captured the shared distress when the parent observed their child experiencing condition-related difficulties. These findings are in line with the findings of the present study.

A South African qualitative study of 79 mothers of children with cleft lip and palate found that mothers reported feelings of alienation from their community, after their friends distanced themselves after the birth of their child. (Hlongwa and Rispel, 2018). These parents also reported feeling shame about other people's reactions to their child's condition. However, this paper only interviewed mothers of young children (*M* age = 3.8 years), so these findings may not be generalisable to parents and caregivers of older children. In addition, research exploring experiences of caring for a child with a long-term health condition has found that parent awareness of their child being bullied can have a negative impact on parent wellbeing. A survey of 251 families of children with a food allergy found that parents who were aware of bullying, scored higher on distress scales and lower quality of life. (Shemesh et al., 2013). The findings of the present study and the existing literature highlight that teasing can have a detrimental psychosocial impact on both young people and their caregivers. The present study is novel in identifying a positive association between parent-reported teasing and parent negative affect and stress in a visible difference population.

Existing research has indicated that the negative attitudes of other children towards appearances that differ from the norm can begin early in life, which may result in stigma or discrimination directed towards the child with the visible difference. For example, a study with 396 primary school students, found that children had significantly less positive attitudes towards a character with a facial burn, compared to a character with no appearance difference (Parnell et al., 2021). It was found that these stigmatising attitudes towards the character with a facial difference develops at age 6-8 years old. Therefore, children of age 6 and above with a visible difference may experience teasing and social exclusion from their peers. However, this study only included a character with a visible facial difference, so these findings may not be indicative of children's attitudes towards individuals with other types of appearance difference. Nevertheless, this supports the finding that parents in the present study are reporting teasing, or anticipation of teasing, when their child reaches primary school age or begins socialising more independently.

Parents have a protective instinct and role in their child's life, consequently when their child is threatened or harmed, they may experience self-doubt in their ability to protect their child (Horridge, et al., 2010). Parents of children with burn injuries have reported being over-protective and controlling in their attempts to cope with low mood and anxiety related to their child's condition (Horridge et al., 2010). As previously discussed (section 2.5.1.), overprotective parenting in parents of children with a visible difference can have negative consequences for child social development (Benson et al., 1991). Therefore, it is essential to support parents in managing their own emotional responses to possible social challenges faced by their child.

Another important finding of this study was the relationship between teasing and positive affect mediated by parent social confidence. In the presence of parent-reported teasing, greater parent confidence in addressing appearance-related social interactions (e.g., managing questions and comments from others), may promote positive affect and may also be protective against stress or negative affect. This is supported by a qualitative study with 11 parents of burn injured children, who identified that gaining confidence in their ability to support their child and their child's ability to take care of themself was an important process in adjustment to their child's injury (Horridge et al., 2010). Drawing on findings from study 1 of this PhD, if parents feel more socially confident, they may also feel more able to equip their child to manage these situations independently. This can help them manage stress and anxiety about challenges their child may face. Therefore, promoting parental social confidence in these situations could be an important target for intervention.

It is important to acknowledge that both the quantitative and qualitative data only reflected parent perceptions of teasing. Consequently, the frequency and impact of teasing reported in this study may not accurately reflect the level of social challenge that the child is currently experiencing. As a result, this data will only capture teasing and bullying that the parent is aware of. Existing literature has found that there are differences between the amount of bullying reported by parents and children (Shemesh et al., 2013). A quantitative study of children with a food allergy found that 45.5% of children reported experiencing bullying, whereas only 36.3% of parents reported that their child had been bullied. In addition, this survey found that 22% of children did not tell their parents that they had been bullied. This suggests that amongst the present parent sample there are likely to be parents whose children have not disclosed teasing and bullying to them. There is also evidence to suggest that older children may be less likely to report bullying to their parents. A large multi-site study of bullying in US schools conducted interviews with 51 students and found that young people did not report bullying for several reasons including: feelings of shame, concerns of parental intrusion, and the belief in parental omniscience (that parents should have known) (deLara et al., 2012). Coupled with the greater autonomy of early adolescence and beyond, a reluctance to disclose teasing and bullying may mean that parents of adolescent children with a visible difference may not have an accurate understanding of the social challenges faced by their child. Nonetheless, given the focus of the present PhD is on parents rather than the direct impact of social challenges on the child, this measure can have utility for understanding the psychosocial impact of perceived social challenges on parents and how this may affect parental adjustment to their child's visible difference.

It is also important to consider that missing data on the parent-reported teasing variable did account for a percentage of the missing data in the overall data set. Associated qualitative findings indicated that some parents felt their child was too young to be exposed to social situations in which teasing, or bullying might occur, and so did not respond to items on this scale. Nevertheless, even when parents reported in the qualitative data that they felt their child was too young to experience teasing or bullying, they described their anticipatory anxiety of social challenges (e.g., starting school). This indicates that even when a child is young and not yet socialising independently, social challenges of the future still preoccupy parents. The qualitative findings indicated that negative reactions from other people had a negative emotional impact on parents, regardless of their child's age. Future research with parents of children with visible differences could include broader questions about interactions with others to capture the emotional impact on parents before the child is socialising independently.

A final consideration when interpreting these findings relates to the COVID pandemic. Although lockdown measures had lifted to some extent, data collection occurred over the summer of 2020 whilst children were not in school. Due to the lockdown and school closures implemented in March 2020, children would not have attended school for several months at the time of data collection. As a result, children's interaction with peers may have been lessened or different to periods before or after the initial COVID-19 lockdown. Some parents reflected on this reduced social contact as a positive (e.g., less teasing or unwanted attention), however others expressed concerns about the impact that this period of social isolation may have on their child's social and emotional development. They also worried that returning to social environments may now be more challenging, as their child had become used to only interacting with immediate family. Further reflections on the impact of COVID-19 on this PhD can be found in Appendix E.

6.4.3. Self-compassion and psychological flexibility

As hypothesised, the present study found that self-compassion was significantly associated with reduced stress frequency and stress difficulty for parents of children with appearance-affecting conditions and injuries. This finding is aligned with the qualitative themes of study 1, particularly the theme "walking the tightrope". In this theme parents were concerned about doing the "right" thing for their child and the potential negative outcomes that may arise if they failed to correctly strike the balance between providing adequate support and giving their child the space to manage independently. This weight of responsibility and self-blame did not leave room for self-compassion for these parents, and they reported negative consequences for their wellbeing as a result of this perception of their parenting role.

The existing condition-specific visible difference literature has begun to identify the important role of self-compassion for parents and caregivers. A quantitative study of 91 parents and primary caregivers of children with burn injuries found that greater self-compassion predicted fewer depressive symptoms (Hawkins et al., 2019). Self-compassion explained a greater proportion of the variance in the depression outcome than shame, and guilt was found to no longer be a significant predictor once self-compassion was included in the model. It is possible that for caregivers of children with burn injuries, self-compassion could be an important protective factor against psychological distress. It is important to acknowledge that this research was with parents of children with burn injuries only, 8 weeks following the injury. This population has been found to experience high levels of guilt related to the cause of their child's injury (Heath et al., 2018), therefore self-compassion might be particularly useful for these parents. However, these findings still have utility in understanding possible protective factors for parents of children with a range of appearance-affecting conditions and injuries.

Self-compassion has also been highlighted as important for the wellbeing of parents, to help them to cope with challenges (Moreira et al., 2015). Mindful parenting has been found to increase self-compassion, as well as general wellbeing in parents (Gouveia et al., 2016). It has been proposed that self-compassion may help parents develop a calm acceptance and compassionate approach to their child's behaviour, particularly when children express negative emotions (Neff & Faso, 2015). A study of 51 parents of children with autism found that self-compassion was positively associated with life satisfaction and negatively associated with depression and parental stress (Neff & Faso, 2015). Self-compassion in parents has also been found to be positively associated with authoritative parenting (high control and warmth and support in parent-child relationship) and negatively associated with authoritarian (high control without warmth and support) and permissive (low control with warmth and support) parenting (Gouveia et al., 2016). Parenting stress was also positively associated with permissive and authoritarian parenting styles. Therefore, this suggests that self-compassion could be protective against parenting stress and supportive of adaptive parenting styles. The findings of the present study are consistent with these conclusions drawn from research with parents from the general population and demonstrate a similar protective effect against psychological distress in parents of children with a visible difference. This exploration of the role of self-compassion in a cross-condition visible difference parent sample is novel and adds to our understanding of the experiences of parents of children with visible differences.

Another important finding of this research was that self-compassion mediated the relationship between psychological flexibility and stress difficulty. This supports existing research which highlights the relationships between anxiety in family functioning, self-compassion and psychological flexibility. Although conducted with self-report data from 500 young people rather than parents, a study grounded in the Circumplex Model of Family Systems (Olson, 2000) found that higher chaotically enmeshed family functioning (characterised by high control, intrusive parenting styles and lack of flexibility; sometimes referred to as "helicopter parenting") was significantly associated with lower levels of psychological flexibility and self-compassion among young people (Berryhill et al., 2018). Berryhill et al. (2018) also found that psychological flexibility and self-compassion mediated the relationship between chaotic-enmeshment and anxiety for young people. Therefore, both psychological flexibility and self-compassion have been related to increased wellbeing and a reduction in maladaptive family functioning. However, as stated, this finding is based on self-report data from the young people within a family rather than taking a systemic approach to assess how self-compassion and psychological flexibility may promote wellbeing in the whole family system. Nevertheless, in agreement with this, the findings from the mediation analysis in the present study may indicate that self-compassion is an important mechanism in the relationship between psychological flexibility and stress difficulty, whereby parent psychological flexibility is protective against parenting stress when facilitated by parental self-comp. As a result, it is important that both psychosocial constructs are considered as targets for intervention.

6.4.4. Parent-child communication

As predicted, the present study found that good parent-child communication related to their child’s visible difference, predicted reduced negative affect and stress difficulty in parents. Some parents also provided further information about communication with their child within the open-ended qualitative questions. Parents described being cognisant of the need to prepare their child to manage independently and believed this could be achieved by regularly speaking to their child about their condition or injury. This included having conversations about their condition or injury to help their child understand their appearance difference and to aid them in building their own narrative, as well as discussing difference and diversity more broadly. Parents also reported an awareness of the need to provide a good model to their child about how to communicate with others (e.g., friends, teachers, public) about their condition or injury.

Communication is an important aspect of interpersonal and interdependent relationships between family members (Branje et al., 2011). Communication is an integral part of parent-child relationships and influential in decision making (Jackson et al., 1998) and development of independence (Hamill, 1994). Clear and effective parent-child communication has been identified in several psychosocial models of family functioning. The Circumplex Model of Marital and Family Systems (Olson, 2000) identified communication as one of three key dimensions in family functioning. Communication is described as a facilitating dimension which promotes the development of the other two key dimensions: cohesion and flexibility. In addition, communication is one of the six dimensions considered by the McMaster Model of families (Miller et al., 2000). Communication is divided into instrumental (information exchange and problem solving) and affective communication. Consequently, theoretical models of family functioning consistently identify communication as important to maintaining positive familial relationships.

Parent-child communication has also been found to be important in the promotion of effective management of long-term health conditions in children and young people. A meta-analysis of 62 studies examining family functioning and management in children with chronic health conditions found that, among other aspects of family functioning, more positive communication was associated with better medical adherence (Psihogios et al., 2019). Furthermore, in a study exploring the transfer of management from parents to adolescents, children with type 1 diabetes reported a high level of parent-child communication related to management of their condition (Hanna et al., 2009). These authors concluded that parent-adolescent communication may be a facilitator of parental support of children with diabetes, and communication was also related to improved diabetes management. A video-feedback intervention to improve communication between parents and children with a disability resulted in increased parental self-esteem (Lam-Cassetteri et al., 2015). As a result, effective parent-child communication is important for both parent and child outcomes in families with a child with a long-term health condition.

The findings of the present study contribute to the understanding of the impact of parent-child communication on parent affect and stress in parents of children with visible difference. To the authors' knowledge, parent-child communication has not been previously explored in this population and therefore this study makes a novel contribution to the visible difference literature. In addition, this provides evidence to support parent-child communication as a possible target for parenting intervention.

6.4.5. Parent knowledge, self-efficacy, and confidence

As hypothesised, the present study found that perceived parental knowledge of their child's condition or injury and satisfaction with any treatment their child had received was significantly associated with reduced negative affect and reduced stress frequency. The analysis also indicated that parental self-efficacy and social confidence were significantly associated with increased positive affect. These findings highlight the importance of parents feeling informed and confident in their ability to support their child.

This is supported by the literature on patient activation (increased knowledge, skills and confidence in managing and advocating for their child's needs, e.g., Hibbard et al., 2004) and the Chronic Illness Care Model (Bodenheimer et al., 2002), which advocates for the integration of patients and caregivers as members of the care team and the implementation of patient-orientated care. Previous research has found that increased activation in parents of children with autism spectrum disorder was positively correlated with parent satisfaction with treatment, ability to self-manage child difficulties and negatively correlated with parenting stress. Additionally, a systematic review found that parent activation although most interventions demonstrated positive outcomes, evidence was inconsistent for outcomes related to parental self-efficacy and confidence (Mirza et al., 2018). Therefore, although there is some evidence to suggest that knowledge, skills, and satisfaction with care may support parent wellbeing in these populations, further work is required to identify the most salient and beneficial content for inclusion in parent activation interventions.

Parents' knowledge of their child's condition and satisfaction with their treatment has been previously explored in parents of children with CLP. A survey study of 1163 parents of children with CLP found that knowledge of condition and satisfaction with treatment was significantly associated with greater positive life orientation (Stock et al., 2020). The present study is the first to investigate this construct as a predictor of parent affect and stress. The qualitative data within the survey also indicated that during the COVID-19 outbreak and lockdown, parents were concerned about the reduced access to healthcare and less satisfied with the support and information they received. Qualitative research about parent's experience of CLP care during the COVID-19 pandemic found that caregivers experienced anxiety and stress in the period during which access to healthcare and support was reduced (Costa et al., 2021). Furthermore, research exploring the experience of providing care during the COVID-19 pandemic found that clinicians reported significant concerns about the mental health and wellbeing of their patients and their families during this time (McWilliams et al., 2022). Consequently, the experience of COVID-19 may have had a negative impact on knowledge and satisfaction with treatment, as well as parent adjustment in the longer-term. The present findings therefore contribute to the visible difference literature by building on understanding of how increased parent knowledge and satisfaction with their child's care can protect against psychological distress, in a cross-condition population.

Existing parenting interventions for parents of children with appearance-affecting health conditions have been found to improve parent self-efficacy in managing their child's condition. The 'Triple P Parenting Progam' is a multi-session intervention which aims to increase parenting skill and self-efficacy related to child behaviour, parenting practices and family stress (Morawska et al., 2016). A systematic review of 10 intervention studies found moderate evidence for the effectiveness of the 'Triple P Parenting Program' for improving parent self-efficacy and confidence in managing their child's condition (Costa et al., 2020). In addition, this intervention was found to improve parent and family quality of life and reduce parenting stress. This supports the present findings and indicates that parent self-efficacy could be an important target for intervention. The present findings also add to the current literature by exploring the role of self-efficacy in parents of children across a large number of appearance-affecting conditions and demonstrates that this is a cross-condition psychosocial factor for promoting parental positive affect.

6.4.6. Appearance investment and appearance fixing behaviours

The present study found that appearance investment was significantly negatively associated with parent negative affect, whilst appearance fixing behaviours were significantly positively associated with stress difficulty. The findings provide some insight into the role of body image focussed cognitions and behaviours in parents of children whose appearance differs from societal norms. This supports the findings from study 1, particularly the sub theme “Shielding my child”, which reflects on parents’ choice to engage in appearance fixing behaviours to conceal their child’s appearance difference, and the possible implications of these actions.

Unexpectedly, appearance investment in parents was associated with reduced parent negative affect. This conflicts with existing findings related to parent appearance investment and attitudes towards others with visible differences. A survey of 183 parents of children without a visible difference found that increased appearance investment in mothers was associated with negative attitudes towards a child character with a facial burn and a child character in a wheelchair (Parnell, 2021). However, parents in Parnell’s study were asked to make judgements about a digital drawing of a child, and so these findings may not generalise to parents’ attitudes and feelings about their own child’s appearance.

However, the qualitative data from the current survey highlighted that a few parents utilised appearance fixing and grooming behaviours to align themselves with societal appearance ideals, as a coping strategy to manage stress and anxiety. This is supported by the existing literature regarding body image coping strategies (e.g., appearance fixing and behavioural avoidance) and wellbeing. Research conducted both in the general population and with adults with visible differences has found associations between body image evaluation and unhelpful body image coping strategies (avoidance and appearance-fixing) (Choma et al., 2009; Zucchelli et al., 2020). A study of 418 students found that appearance fixing, and avoidant coping were both significantly negatively associated with subjective wellbeing (Choma et al., 2009). It is possible that a bi-directional relationship exists between parent negative affect and appearance investment, indicating that increased appearance investment may act as a buffer against the experience of negative affect.

As predicted, the present study also found that increased parent-reported, child-focused appearance fixing behaviours (e.g., altering their child's appearance to align with societal norms such as, hiding or covering their appearance difference) predicted stress difficulty. This is in line with research that explored relationships between objective severity of an appearance difference and psychosocial adjustment. A study of 400 patients with a range of visible differences found a significant quadratic relationship between objective severity and psychosocial adjustment (Moss, 2005). Moss concluded that because individuals with a very severe difference or a non-severe difference will experience relatively consistent social reactions, they are likely to be able to adjust to these reactions over time. However, if the condition's severity is judged to be in the middle of this spectrum, social reactions will vary more, making interactions unpredictable and more challenging to adjust to. Furthermore, a qualitative study with 15 adults with visible differences found that these individuals experienced anxiety when faced with the prospect of disclosing their visible difference to other people (Sharratt et al., 2020). Therefore, if parents choose to conceal their child's appearance difference, they may not have the opportunity to adjust to consistent reactions. Due to this lack of exposure, parents may remain anxious or stressed when anticipating situations in which their child's condition or injury may become visible to others and invoke unpredictable reactions.

When considering body image focussed cognitions and behaviours in parents, it is important to consider how these may impact their child with a visible difference. Interestingly, Parnell (2021) found that parental attitudes did not predict child attitudes towards a variety of different appearances, including a character with a facial burn. As the present study aimed to investigate whether increased appearance investment would result in greater challenges to affect and stress in parents of children with a visible difference, it may be concluded from the present findings and the existing literature that the mechanism between parent-child body image cognitions and behaviours is not yet fully understood. Further research is required in this area to better understand the role of the parent-child relationship in the development of body image focussed cognitions and behaviours.

6.4.7. Social support

Study 1 findings indicated that parents often perceived social support to be inappropriate or insufficient to meet their needs (e.g., lack of peer-support) which appeared to negatively impact wellbeing. Other research also suggests that parents of children with a visible difference find social support useful (Baker et al., 2009; Bogart et al., 2017). However, contrary to expectations, social support was not a significant predictor in the stress frequency and difficulty models. It is conceivable that the quantitative design of study 2 was limited in its ability to reflect what may be a more nuanced relationship between support and wellbeing, that also considers the nature of support and whether it meets parents' needs, as indicated by the qualitative data. In line with this, a survey of 568 women with fibromyalgia syndrome found that perceived quality of social support was associated with lower levels of depression, higher levels of self-efficacy, and greater overall psychological wellbeing, whereas larger social support networks were only associated with greater pain and symptom management (Franks et al., 2004). Consequently, these authors concluded that quality of social support was more important than quantity. To better understand the role of social support for this parent population, future research should consider measuring quality and appropriateness of social support rather than simply the presence or absence of it. Furthermore, the qualitative data indicated that due to the COVID-19 lockdown, many individuals had much reduced access to social support. This included information support (e.g., friends and family) and more formalised support (e.g., charity support groups). Reduced social support may have had a negative impact on parent adjustment during this time. This has important implications for understanding the experiences of parents and how to better meet the support needs during any future periods of similar social isolation.

6.4.8. Child development

As predicted, child age was significantly negatively associated with both parent negative affect and positive affect. The relationship between child age and negative affect supports the study 1 findings and indicates that parents found adjustment to their child's visible difference the most challenging in the early years of life. Although this does assume that children with acquired conditions or injuries are developing an appearance difference when they are younger, which may not be the case for all. However, the relationship between child age and positive affect is not in line with predictions based on study 1.

Literature addressing the broader concept of coping and psychosocial adjustment states that although most work focuses on the experience of negative affect, positive affect can co-occur during times of chronic stress (Folkman & Moskowitz, 2000). It is possible that these findings reflect the simultaneous experience of both positive and negative affect in parents as their child develops and encounters new experiences and challenges. For example, a parent of child with a visible difference may be proud and excited that their child is starting secondary school, however they may also feel anxious about their child's increasing social independence. These findings highlight the complexity of the parenting role and the ongoing challenges to psychosocial adjustment for parents of children with visible difference.

The majority of the existing literature has been conducted with young children or pre-adolescents. Therefore, these findings contribute to our understanding of the experience of parents with older children in mid-late adolescence, and how parent psychosocial adjustment continues to fluctuate with child development. Consequently, support for parents of children with visible differences should be available to parents of children at all ages, and not just focussed on infants and younger children.

6.5. Limitations

Whilst this study provided novel insights into risk and protective factors for parents of children with visible differences, it is important to recognise limitations. Although the study sample was more diverse than the sample of study 1, there are still limitations in the generalisability of the findings. Despite efforts to broaden the representation of different family structures, most participants were from ‘traditional’ two parent families, thus the present findings may not apply to families comprised of non-parent carers or single parents. The broader paediatric literature has noted that the experience of single parents of children with chronic health conditions is currently underserved by research (Brown et al., 2008; Granek et al., 2014). The implications of lack of representation within research is discussed further in section 9.2.1.1.

As previously discussed in section 2.8., a common limitation of literature within this field, is the underrepresentation of fathers and male caregivers. Similarly, a gender imbalance was present in the current sample with most participants being mothers. As previously noted, (section 2.8.) previous research has found that there are differences in variables which predict adjustment in mothers and fathers of children with CLP (Stock et al., 2020), suggesting that female and male caregivers may have different experiences and support needs. As a result, the results of the present study may not generalise to caregivers of all genders and roles.

The research design was focussed on exploring associations between psychosocial constructs and psychological wellbeing outcomes, it is not possible to determine cause and effect from this design. In addition, the regression models only predicted between 23-33 percent of variance in the psychological wellbeing outcomes, a large proportion of the variance remains unexplained., Although this study adds important new knowledge to the field of visible difference, it does not provide a full understanding of the factors which affect the psychosocial adjustment of parents of children with visible differences.

Several of the measures were adapted or items written specifically to meet the aims of the present study. As discussed in section 6.2.2.1., these measures did not have established or published psychometrics to indicate their reliability or validity. Although all study-specific measures underwent a feedback process via the PAG to ensure relevance and appropriateness, the adaptation and inclusion of study-specific measures may limit generalisability of the findings to samples beyond this study. All measures were also self-report and completed by parents. Whilst this facilitates efficient and replicable data collection, the findings are limited to what parents can, wish, or feel able to report. As discussed above (section 6.4.2.), this can be problematic for variables such as teasing. Children may not report teasing to their parents (Shemesh et al., 2013), so parent-report may not be an accurate representation of social challenges experienced. Therefore, parent-report measures can limit our understanding of the parent and child experiences of living with a visible difference and impact generalisability of the findings.

Finally, this study did not employ a comparison group to explore the differences in adjustment between parents of children with a visible difference and parents of children in the general population. Previous research has found that parents of children with CLP and infantile haemangioma (a red birthmark) report less distress and stress than parents of children without a visible difference (Van Dalen et al., 2021a). These findings suggest differences between the two parent populations. As this comparison is not available within the present study, it may not be possible to conclude which aspects of the findings are specific to parents of children with a visible difference, rather than a result of the typical challenges of parenting.

6.6. Conclusion

The present study aimed to identify cross-condition risk and protective factors for parents of children with a visible difference. Parent-reported teasing was found to be a risk factor for increased negative affect and stress. Self-compassion, good parent-child communication, knowledge of condition and satisfaction with treatment appear to be protective factors against the experience of parent distress and stress. Self-efficacy and social confidence were found to be associated with increased positive affect. The analyses also highlighted the important mediating role of self-compassion in the relationship between psychological flexibility and stress difficulty. In addition, social confidence was found to mediate the relationship between parent-reported teasing and positive affect. The results also indicated that further research is required to explore the complex relationship between parent body image cognitions and behaviour, and the impact this may have on their child with a visible difference. Finally, results suggest that parents may continue to experience a combination of positive and negative affect as their child grows and develops, in response to continuously evolving parenting experiences and challenges. This emphasises the importance of support for parents throughout their child’s development. This study adds to the findings of study 1 and identifies possible targets for the development of psychosocial support for parents. The following chapter addresses the integration of the data sets from studies 1 and 2 and discusses overall conclusions.

Chapter seven: Mixed methods integration

7.1. Introduction

This mixed methods PhD employed an exploratory sequential design (see section 3.6.1. for detail about this design). Data analysis and interpretation occurred at several points in this exploratory design (Creswell & Clark, 2017). Initially, parent experiences were explored using a small sample and a qualitative design. Based on these findings, a larger quantitative study was designed and conducted. Data were analysed first following the qualitative data collection (see chapter 4), then mapped onto a quantitative design (see chapter 5), and finally the quantitative data were collected and interpreted (see chapter 6). Following these stages, the interpretations drawn from the qualitative and quantitative data sets were integrated to form overarching mixed method conclusions (Creswell & Clark, 2017). This chapter will detail the process and outcomes of this integration and discuss the rationale for the approach selected to integrate these data sets. Finally, the chapter will discuss the overall conclusions presented in the context of relevant literature and the aims of the PhD.

7.2. Overview of findings

Before discussing the method of integration and overall conclusions, this section will briefly present the key findings from studies 1 and 2.

7.2.1. Overview of qualitative study (study 1)

Twenty parents and 11 health and support professionals took part in one-to-one telephone interviews and 25 parents participated in focus group discussions at a charity event. Interviews and focus groups explored the experience of caring for a child with a visible difference, support available to parents, and suggestions and priorities for future support. Professionals reflected on the common concerns brought to them by parents and carers, existing parent interventions, and what they felt future support should include. Across this sample, 10 different appearance-affecting conditions and injuries were represented.

Data from the three data sets (parent interviews, professional interviews, and parent focus groups) were integrated using a triangulation protocol (See section 4.2.7.). Three main cross-condition themes were identified: 1) Appearance does(n't) matter, 2) Being "battle" ready, and 3) Walking the tightrope.

"Appearance does(n't) matter" described the psychological challenges of being a parent of a child with a visible difference. Many described the difficult thoughts and feelings (e.g., anxiety, guilt, shock, and low mood) that they experienced in response to their child's visible difference. These were particularly prevalent in the early days of their child's condition or injury, either in the first months of life or immediately following the injury or illness event which had caused their visible difference. Parents also spoke about rebuilding from this difficult point, as their child grew and developed. For many, as time passed, their child's personality, hobbies, and interests became their defining attributes, whilst their visible difference faded into the background of life.

"Being 'battle' ready" captures parents' worries about the challenges their child may encounter. Parents were particularly preoccupied with difficulties related to social interactions and interpersonal relationships. Some spoke about the desire to shield their child from social challenges, typically by hiding or concealing their child's visible difference. Professionals reflected that this instinct seemed to be a coping strategy for parents, rather than being beneficial for the child themselves. Many parents spoke about their motivation to properly equip their child with skills and resources (e.g., confidence and resilience) to manage challenges independently. However, many also felt unsure about how best to provide this support to their child.

“Walking the tightrope” described parents’ struggle with the lack of clarity around how to best support their child. They spoke about the difficulty of striking a balance between providing enough structure and support, whilst not making the child’s visible difference the whole focus of their relationship. Parents discussed their concerns about the possible negative consequences if they were to fail to strike this balance correctly. Many were worried about how their child would cope if they struggled to manage challenges independently and how they would cope if they saw their child struggling.

7.2.2. Overview of quantitative survey study (study 2)

In this study, 209 parents and carers of children with a visible difference completed an online survey which included standardised and study-specific measures and open-ended questions asking about their experience of caring for a child with a visible difference. This survey also facilitated the investigation of possible risk and protective factors for psychological distress. The psychosocial constructs that were assessed in this survey were identified in the qualitative data and mapped onto quantitative measures (See chapter 5) The qualitative data collected from open-ended questions enhanced and clarified aspects of the quantitative data (See section 6.2.7.6.).

No significant differences were found between parents of children with congenital and acquired conditions on any outcome variable. This supports the cross-condition themes identified in the qualitative data in study 1. Increased parent-reported teasing was significantly positively associated with negative affect and stress. Increased knowledge of condition and satisfaction with treatment and good parent-child communication were both significantly negatively associated with negative affect and stress. Parental social confidence and self-efficacy were both significantly positively associated with positive affect. Finally, self-compassion significantly mediated the relationship between psychological flexibility and parent stress difficulty.

The qualitative data indicated that some parents struggled with the emotional impact of managing other people's reactions to their child's condition or injury. Parents also reflected upon their desire to prepare their child to manage these social situations independently. They reported a focus on providing their child with a positive narrative, conversations about difference and diversity, and modelling adaptive responses to challenging social situations (e.g., unwanted comments or questions). Finally, parents reported their experiences of the COVID-19 pandemic and their associated concerns. Parents were worried about the impact of social isolation on their child's social and emotional development. They also stated concerns about reduced access to healthcare services and support, which led them to feeling less knowledgeable about and satisfied with their child's care.

7.3. Methods of integration

Integration in mixed methods research has been defined as "the interaction or conversation between qualitative and quantitative components of a study" (O'Cathain et al., 2010, p.1147). The integration of findings from data sets is an essential element of conducting mixed methods research (Creswell et al., 2004). The following section will outline established techniques of mixed methods integration and explore the method selected to integrate the qualitative and quantitative elements.

7.3.1. Three techniques for integration

The three techniques for integrating mixed methods are: 1) triangulation protocol, 2) "following a thread", and 3) mixed methods matrix (O'Cathain et al., 2010). Two of these were utilised previously (see chapter 4 for triangulation and chapter 6 for 'following a thread'). Briefly, triangulation is used when a researcher has employed qualitative and quantitative methods to examine different elements of the same research question (O'Cathain et al., 2010). Integration using this method is conducted at the interpretation stage of the research, when both the qualitative and quantitative components of the research have been analysed separately (See Figure 18). The findings for each component are then listed and compared to determine the level of agreement between data sets (Farmer et al., 2006). "Following a thread" is described as using one data set to further explore or clarify an aspect of another (Moran-Ellis et al., 2006). Although there is no standardised protocol for this method, it typically involves identifying a theme or particular question in one data set and following it across to the other data set to enhance understanding. Finally, the mixed method matrix technique can be used when both qualitative and quantitative data are available from the same case (e.g., the same participant; Bazeley, 2009). In this situation all the data available for a single participant are analysed together, focusing on individual participants rather than broader examinations of variables or themes (O'Cathain et al., 2010).

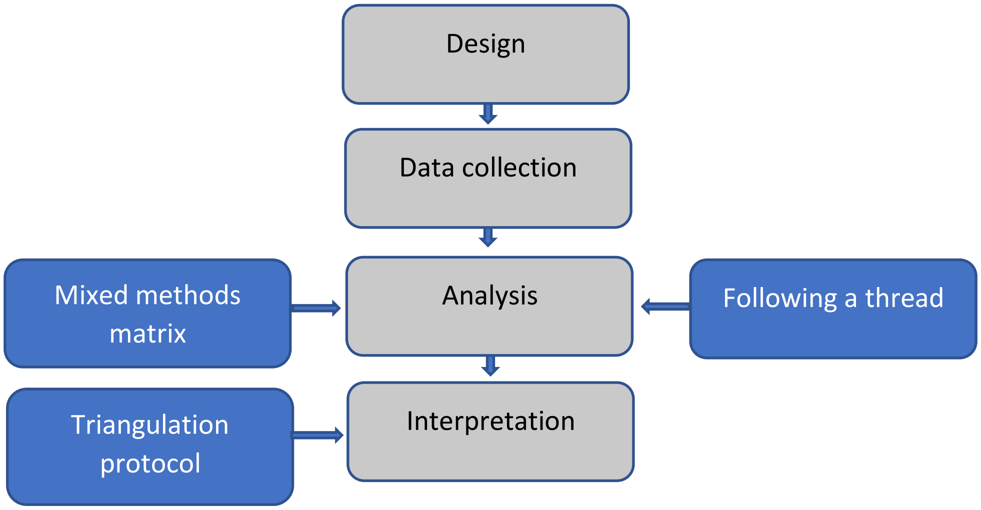


Figure 18: Diagram of stage of application of each technique of mixed methods integration (O'Cathain et al., 2010)

7.3.2. Triangulation protocol

To combine the qualitative (study 1) and quantitative (study 2) elements, a triangulation protocol was selected. As stated above (section 7.3.1.), this integration process occurs after both the components of the mixed methods research have been analysed independently. There are several methods of triangulation: methodological, data, and investigator. Methodological triangulation describes the process of integrating findings from more than one research method (Farmer et al., 2006). Although originally designed for integrating data from multiple qualitative methods, this method has been utilised in mixed methods research and found to be relevant for mixing qualitative and quantitative data sets (O'Cathain et al., 2010). The most detailed triangulation protocol, published by Farmer et al. (2006), utilises a "convergence coding matrix" where findings from each component are displayed side by side and then reviewed to determine whether there is agreement, partial agreement, silence, or dissonance between the data sets. The level of agreement across data sets is then assessed to determine where the two data sets agree, add to, or contradict, and a summary of the combined findings is then created. The process of integrating data using this method is illustrated in Figure 19.

The triangulation protocol has several advantages which make it the most appropriate approach for the present mixed methods research. Firstly, it is the only method to acknowledge and explore silence. Silence occurs when a topic or theme is found in one data set but not in another (O'Cathain et al., 2010). Silence is important to consider as it can highlight the way in which different methods have explored different facets or perspectives of the topic. This can facilitate a more in-depth understanding of a phenomena by identifying a broader range of voices and concepts (O'Cathain et al., 2010). The triangulation protocol also requires that researchers move from thinking about findings from each method, to thinking about "meta themes" which span multiple data sets (Farmer et al., 2006). This is an important step to pragmatic intervention development, a primary aim of the present PhD. Exploring meta-themes within the mixed methods data were essential in the process of identifying constructs which should form the core components of a parenting intervention. Finally, the triangulation protocol provided the most detailed and clear account of how to approach a mixed method integration (Farmer et al., 2006b). A primary critique of mixed methods research is the lack of clarity around the specific methodological steps taken when integrating findings from multiple data sets (Feilzer, 2010b). Therefore, it is often unclear whether true integration has been achieved. Utilising a method with a clear and replicable protocol demonstrates rigour and transparency in the integration process.

The integration of the study 1 and study 2 data sets was conducted using the convergence coding matrix method outlined above and illustrated in Figure 19. As the present PhD adopted an exploratory sequential mixed methods model (see section 3.6.1.), the themes generated in study 1 acted as a guide for the integration. All main themes and sub-themes from study 1 were examined and the findings relating to the corresponding psychosocial constructs were summarised and compared. Based on this comparison, the PhD student assessed the level of agreement of the findings of the two data sets (e.g., agreement, partial agreement, disagreement, or silence). An interpretation of the mixed methods findings was then made based on the two sets of findings and the level of agreement. The full joint display table illustrating this process can be found in Appendix I.

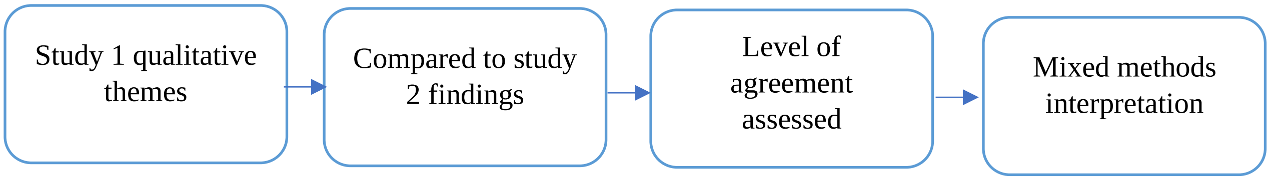


Figure 19: Illustration of the systematic process of integrating study 1 and study 2 data sets.

7.4. Learnings from the mixed methods integration

A joint display table was used to present the mixed methodinterpretations (Creswell & Clark, 2017). Overall, the integration indicated a high level of agreement between the two data sets. Study 1 and study 2 data demonstrated agreement on stress and distress being present for parents, at some stage in their child’s journey. There was also agreement about parent concerns related to social challenges, their desire to protect or shield their child, and the drive to prepare their child to manage oncoming challenges. Both data sets also demonstrated that parents lacked confidence in how best to support their child and that certain psychosocial resources (e.g., self-compassion, psychological flexibility, social support, and perceived knowledge) can help parents to manage their own thoughts and feelings. There were some disagreements in the data set related to what age parents seem to find these difficulties most challenging. In addition, only study 1 indicated that there may be differences between the experiences of parents of boys and girls. Finally, there were several topics in which there was silence on one of the data sets, including all data related to COVID-19. The full joint display table can be seen in Appendix I. The following section will describe the findings and reflections in greater detail.

7.4.1. Parent psychological distress

Both data sets indicated that some parents experience psychological distress related to their child’s visible difference. Several challenging feelings identified by parents were anxiety, low mood, guilt, and loss. There was partial agreement between the data sets with regards to the relationship between child age and parent negative affect and stress. The qualitative findings indicated that difficult thoughts and feelings were most challenging in the early days of having a child with an appearance-affecting condition or injury. This agrees with the quantitative study, which found a significant negative relationship between child age and negative affect. However, the quantitative study also found a significant negative relationship between child age and positive affect. A possible interpretation of these integrated findings could be related to ongoing and shifting challenges that arise as a child gets older. As a child grows up, different transition points and challenges may arise and trigger a variety of difficult and challenging emotions in parents. For example, an interview study of parents of children with craniofacial conditions found that parents were anxious about their child starting a new school, as this social transition would result in their child entering a new environment with unfamiliar children (Feragen et al., 2021). Therefore, new transition points in the growing child's life could result in challenges to adjustment for their caregivers.

In the qualitative data, many parents also highlighted that as their child developed cognitively and emotionally, other aspects of their identity become more salient and important than their appearance (e.g., their personalities, hobbies, talents etc.). As a result, their visible difference became less relevant to parents and triggered fewer psychological challenges for them (Thornton et al., 2021). This highlights the complexities and ongoing changes and challenges in the life of a parent of a child with a visible difference.

The qualitative data in study 1 indicated that there may be differences between the experiences of parents of children of different genders. However, this finding was not echoed in the study 2 findings and child gender was not a significant predictor of parent distress or stress. Within study 2, parents did not report high levels of appearance investment. Therefore, these parents may not value appearance as an attribute and so may be less preoccupied by the gender differences in appearance ideals.

7.4.2. Preparing their child to manage challenges independently

The mixed methods integration found a high level of agreement between data sets regarding parental concerns about social challenges related to their child's visible difference. Both data sets also explored parents' approach to managing the threat of social challenges. The quantitative findings indicated a significant positive relationship between parent-reported noticeability of their child's visible difference and stress frequency. This suggests that parents who find their child's condition noticeable may experience stress related to the visible difference more often. There was a high level of agreement between data sets regarding parents' desire to shield their child from unwanted social interactions and attention. Both data sets found that parents may employ appearance-fixing behaviours to negate their own concerns about their child's appearance difference and the possible impact of social challenges. There was also a significant positive relationship between appearance-fixing behaviours and stress difficulty, which suggests that these concealing or fixing behaviours were not effective in ameliorating stress and may contribute to psychological challenges for parents.

There was agreement across the data sets that parents also wish to equip their child with the psychosocial tools to independently manage challenges related to their visible difference. It was clear from both data sets that parents wanted to be able to communicate openly with their child about their appearance difference, as well as diversity in human characteristics more broadly. This open parent-child communication was significantly negatively associated with negative affect and stress in parents. Both data sets identified the importance of parents modelling adaptive responses to social challenges, so their child would be able to observe and imitate these responses and manage situations independently. The quantitative data analysis found a significant positive relationship between social confidence and positive affect. This suggests a relationship between parental social confidence and positive adjustment to caring for a child with a visible difference.

7.2.3. How to provide support

The mixed methods integration also found high levels of agreement across data sets when exploring parents' feelings about supporting their child. The qualitative and quantitative data found that some parents struggle with a lack of confidence and clarity about providing support. In addition, the quantitative findings indicated a significant positive relationship between self-reported self-efficacy and positive affect. This is in line with the qualitative findings which suggested that parents who feel more confident about their ability to support their child, may adjust better in their caregiver role. Striking the correct balance around appearance talk and discussing the child's visible difference was a key area of concern highlighted in the qualitative data. As discussed above, agreement with this was found in the quantitative data which indicated that effective parent-child communication may be protective against psychological distress and support psychosocial adjustment in parents.

Both data sets also agreed upon the experiences of parents who reach out to external sources for guidance on how to support their child and that feeling informed about their child's condition or injury is important in promoting parenting wellbeing. A significant negative relationship between knowledge and satisfaction with treatment and parent negative affect was apparent in the quantitative data set. This suggests that these factors may be protective against psychological distress. However, mixed experiences with both informal social support and support from health professionals were identified within both the qualitative and quantitative data. The mixed methods survey data also indicated that the quality of social support was equally, if not more, important than quantity of support in protecting psychosocial wellbeing

The second study found that COVID-19 had a negative impact on parent satisfaction with their child's care due to a reduction in in-person support and cancellations of non-urgent appointments and procedures during the initial wave of the pandemic. There was silence on this topic within the other data, due to the timing of study design and data collection (study 1 conducted in 2019, prior to the pandemic).

7.4.4. Consequences of “getting it wrong”

The qualitative and quantitative data highlighted parent concerns about the possible negative impact of their child’s visible difference and the consequences that may arise if they did not provide adequate support. Across both data sets parents reported guilt and anxiety if their child struggled to adjust and/or had concerns about what would happen if they struggled in the future. More specifically, the qualitative data highlighted parental concerns about their own ability to cope if they witnessed their child struggling. There was agreement across data sets in how certain approaches could be effective for parents to manage these challenging thoughts and feelings. Parents who were able to sit with these thoughts and emotions and practice self-compassion were less likely to experience stress related to their parenting role. The quantitative data set found that self-compassion mediated the relationship between psychological flexibility and stress difficulty. Therefore, both self-compassion and psychological flexibility may be key protective factors for parental wellbeing.

Overall, the triangulation protocol found a high level of agreement between the two data sets. There were several areas where the methodologies were able to highlight different facets within the data, providing a more comprehensive understanding of the phenomena. The following sections will reflect on the integration process and draw on these integrated findings in the context of existing literature and explore implications for intervention development.

7.4.5. Reflexive analysis of integration process

Conducting the integration process of study 1 and study 2 presented multiple challenges. As described above (section 7.3.2.), the qualitative themes were used as a guide for the integration. Given my previous experience with qualitative methods and clinical work, using the participants own words and experiences to guide this phase of the research felt like a comfortable and intuitive process. This bolstered my confidence conducting the integration of qualitative and quantitative findings, which was not something I had prior experience of. I have previously used the triangulation protocol in the PhD for integrating qualitative findings (see 4.2.7.), so this was a familiar process which enable me to focus on the content of the integration rather than learning the process.

One element of the process which I reflected most upon was the presence of silence across either the qualitative or quantitative data sets on certain topics. Much of the silence between the two data sets was as a result of study 1 being conducted in 2019 (pre-COVID-19 pandemic) and 2020 (mid COVID-19 pandemic). This led me to consider the extent to which generalisability of research findings are dependent on social, economic, and political contexts. This challenged my assumptions of generalisability and transferability of findings between different social contexts and further reinforced the importance of including diverse samples and considering intersectionality.

Although the integration process showed a high level of agreement between data sets, I was cognisant that there were limitations to the overall generalisability of the mixed method conclusions. The specific limitations, such as sample characteristics, and the implications of these will be described in detail below (section 7.5.4.). However, the integration process provided me with the opportunity to reflect upon and consider which voices in the parent community were being represented in the mixed methods findings and who had not been included (e.g., lack of representation of male carers, individuals from different cultural backgrounds).

7.5. Discussion

The aims of the mixed methods research were to: 1) explore the cross-condition experiences of parents of children with a visible difference, 2) identify risk and protective factors for psychosocial adjustment in parents of children with a visible difference. The mixed methods integration identified that many parents experience psychological distress, regardless of the cause or nature of their child’s visible difference. They worry about upcoming challenges related to their child’s visible difference and are highly motivated to protect their child from any negative consequences, but feel ill equipped to manage their own reactions to their child’s visible difference and are unsure about how best to provide effective support to their child. These findings highlight a clear gap in psychosocial support for parents and carers. The implications of these findings will now be discussed alongside existing literature and theory, with an aim of intervention development to address this gap.

7.5.1. Self-compassion and psychological flexibility

A key finding of the integrated data was the role of psychological flexibility and self-compassion as a protective factor against psychological distress. This is in line with existing literature exploring adjustment in adults with a visible difference. A quantitative survey study with 78 burn injury patients found that appearance anxiety was negatively related to reduced acceptance of distressing emotions, less engagement with committed action, and reduced ability to defuse from negative thoughts, all elements of psychological flexibility (Shepherd et al., 2019b). These authors suggested that this provides preliminary evidence for the role of psychological **in**flexibility in the maintenance of appearance anxiety in individuals with a visible difference. Self-compassion has also been found to play an important role in the wellbeing of individuals with a visible difference. A randomised pilot trial of a self-compassion writing intervention for individuals with visible skin conditions found reduced negative affect in the intervention group, compared to the control group (Sherman et al., 2019). Furthermore, a quantitative study of 91 caregivers of burn-injured children found that individuals who reported higher self-compassion also reported fewer symptoms of depression and post-traumatic stress syndrome (Hawkins et al., 2019). Although, research has begun to examine the role of these constructs in parents, evidence regarding the role of psychological flexibility for parents remains scarce. In this respect, the present PhD provides a novel contribution to the literature and is the first to explore the cross-condition role of psychological flexibility and self-compassion in parents of children with a visible difference.

Both psychological flexibility and self-compassion are closely linked to the Acceptance and Commitment Therapy (ACT) therapeutic model (Hayes et al., 2006c; Luoma & Platt, 2015; Marshall & Brockman, 2016). The ACT model contains six core processes all of which have the shared goal of increasing psychological flexibility (Hayes et al., 2006). These six processes are: 1) acceptance, 2) cognitive defusion, 3) being present, 4) self as context, 5) values, and 6) committed action. These processes and their relevance in psychological interventions for visible difference will be discussed in greater detail in Chapter 8 (section 8.1.1.).

Although self-compassion is not an explicit component of ACT, there is documented theoretical overlap between self-compassion and psychological flexibility (Marshall & Brockman, 2016). Empirical evidence from a survey study of 144 university students found that self-compassion is significantly correlated with psychological flexibility processes (Marshall & Brockman, 2016). Marshall and Brockman (2016) found that self-compassion and psychological flexibility significantly predicted variance on several wellbeing measures including depression, stress, and life satisfaction. In the present study, self-compassion mediated the relationship between psychological flexibility and stress difficulty, providing further support for the theoretical association between psychological flexibility and self-compassion. The present study is novel in its approach of investigating the connection between these concepts in a population of parents of children with visible differences.

As the present PhD has demonstrated that both these psychosocial constructs are important in supporting the wellbeing of parents of children with a visible difference, a parenting intervention based on the core principles of ACT could be an appropriate next step for support development. This will be discussed further in the following chapter.

7.5.2. Parent-child communication

Parent-child communication was also an important protective factor against psychological distress. Qualitative research with 18 parents of children with a range of visible differences has also found that some parents can find appearance-focussed conversation difficult, (Zelihić et al., 2021). Parents reported finding it challenging to identify the appropriate setting and feared contributing to appearance concerns and upsetting their child. Furthermore, in a qualitative study of 33 parents of children with craniofacial conditions, almost all participants reported concerns about creating a negative awareness of their condition or distressing their child (Feragen et al., 2021). Parents also acknowledged that this conversation would likely become unavoidable and described an ongoing lack of confidence about how to address it led to challenging emotions. Some parents reported that they chose to make room for discussions of difference and diversity more broadly (Feragen et al., 2021). The findings described by Zelihić et al. and Feragen et al. support the integrated mixed methods data of this PhD. This suggests that clarity and confidence around open parent-child communication is important for parent wellbeing. The present PhD also builds on the current literature by presenting empirical evidence of the protective effect of parent-child communication against psychological distress in this population.

Given the evidence from the mixed methods findings and the existing literature, parent-child communication could be an appropriate target for psychosocial intervention. Content related to parent-child communication could be presented within an ACT framework (e.g., using skills taught in ACT to facilitate value-based action and improve parent-child communication). This could encourage parents to be open and adaptive in their approach to communication and support them to manage difficult thoughts and feelings about challenging conversations. In addition, themes of self-compassion could also help parents to cope with difficult thoughts and feelings related to their ability to successfully support their children through these conversations.

Additionally, a self-compassionate approach could provide parents with tools to manage their own worries about appearance-related conversations. As the present PhD has demonstrated that this psychosocial construct is important to supporting the wellbeing of parents of children with a visible difference, content to address parent-child communication will be included in the development of support materials.

7.5.3. Seeking information and support

The present PhD found that although parents were eager to receive information and support related to their child's condition or injury, they often had mixed experiences when they did reach out for support. Some parents felt dismissed by health professionals and others found that the social support offered did not always meet their needs. The second study of this PhD added to this understanding, by providing empirical evidence of the possible protective role of feeling informed and satisfied with care in relation to parent psychological distress. The quantitative data also demonstrated that the mere presence of social support is not sufficient to be protective against psychological distress. Quality and appropriateness of support appear to be the more important factors.

Existing research with parents of children with a visible difference has found that knowledge and satisfaction with care can be beneficial for parental wellbeing. A questionnaire study with 1163 parents of children with CLP found that knowledge and satisfaction with care was protective against psychological distress in both mothers and fathers (Stock et al., 2020). Recent qualitative research has also recommended that, given parents experience their own emotional challenges related to their child's condition or injury, it should be the health professionals' role to support parents with these difficult feelings (Feragen et al., 2021). Therefore, it is important that parents feel equipped to seek the support and information they need from health care services.

Further informal varieties and sources of social support have also been found to be important in parental wellbeing. An interview study with 13 parents and carers of burn injured children found that caregivers experienced a predominant feeling of emotional and physical isolation (Heath et al., 2018a). This study found that peer support, from individuals who have similar experiential knowledge and understanding, is important to parents. Other parents in the same sample reported difficulties seeking support from others who did not have the same lived experience (e.g., health and support professionals, Heath et al., 2018a). This further emphasises the importance of parents accessing social support that is in line with their psychosocial needs.

The findings and literature discussed suggest that informational and emotional support from others could be a valuable protective factor against psychosocial distress. Notably, it is the nature and quality of the support received that is most influential in promoting parental wellbeing. Therefore, equipping parents with the skills and knowledge needed to access appropriate support could be a target for intervention development. This content could also be presented within an ACT framework. Aspects of self-compassion and psychological flexibility (e.g., value-based action) could be important tools to help parents manage difficult thoughts and feelings that they may encounter when seeking and accessing support. A self-compassionate approach could also be useful for parents when identifying and prioritising their own support needs.

7.5.4. Limitations of mixed methods integration

Although the conclusions drawn from the mixed method integration have important and novel implications for the understanding of parent experiences and future support development, the limitations of the research must be acknowledged. Firstly, the samples of both studies are limited in terms of the diversity they represent. Despite attempting to recruit from a wide range of backgrounds, the samples were comprised mostly of individuals who identified as White British. Although this is not uncommon in psychological research (Muthukrishna et al., 2020), it is valuable to reflect on the implications. Parents of children from different cultures hold different health beliefs and coping strategies related to paediatric chronic illness (e.g., childhood cancer, Banerjee et al., 2011), consequently it may not be possible to generalise these conclusions beyond a White British sample. Furthermore, the majority of participants in both studies were mothers (>70% in all samples). Although a gender/role imbalance in the favour of women/mothers is not unusual in parenting and health research (Davison et al., 2017), existing literature with parents of children with CLP has highlighted difference in the experiences and coping strategies of mothers and fathers (Stock et al., 2020; Stock & Rumsey, 2015). As a result, it is important to consider the generalisability of the conclusions drawn from this mixed methods integration. Although the studies attempted to recruit non-parent caregivers, few were recruited to either study. It is therefore possible that these findings cannot be generalised to all children with a visible difference.

To understand the strength of the overall conclusions it is also vital to consider any limitations in the method of integration utilised. Some authors have critiqued the use of parallel or coordinated mixed methods designs (wherein results are combined after each study is completed, as in the present PhD), suggesting that coordinated designs do not reflect the ‘true integration’ of combining methods throughout the research process, as is done in an integrated design (Bryman et al., 2008). However, coordinated designs are considered an appropriate approach for a pragmatic epistemological framework (Greene et al., 2001), as they allow one set of data to enhance or refine the other, highlighting different aspects of the data and providing a basis to draw multifaceted overarching conclusions. Therefore, although integration occurred following the completion of the two studies, the process was rigorous and in line with the aims and theoretical position of the PhD.

7.6. Conclusion

The mixed methods integration of the first two studies of this PhD found a high level of agreement between data sets. The two data sets worked to enhance, clarify, and present alternative perspectives on parents' experiences, coping strategies, and support needs. The integrated data sets highlighted that psychological flexibility and self-compassion were important in protecting against psychological distress. Therefore, an ACT model could be utilised as the basis of a psychosocial intervention for parents of children with a visible difference. There were other psychosocial factors that were highlighted by the integrated findings: anticipation of social challenges, parent-child communication and modelling, and gaining appropriate knowledge and support. The findings provide important evidence that these could be targets for intervention. To the author's knowledge, this is the first mixed methods investigation of the experience of parents and carers of children with a visible difference. Consequently, these findings also make an important contribution to the existing literature and the understanding of parent experiences and support needs.

Chapter eight: Developing an intervention for parents of children with a visible difference

8.1. Introduction

The mixed methods integration of study 1 and 2 highlighted key targets for intervention. Psychological flexibility and self-compassion were important protective factors against psychological distress in parents of children with a visible difference. Parent-child communication and modelling, as well as gaining appropriate knowledge and support, were also identified as protective factors against psychological distress. Actual and anticipated social challenges were identified as risk factors for psychological distress in parents. Based on these constructs, the present PhD aimed to develop a psychosocial intervention for parents. This chapter will address the intervention design and development process. Firstly, the theoretical background of the intervention components will be explored in detail. This will be followed by a discussion of the process and procedures of iterative intervention development, including the use of a collaborative Participatory Action Research approach. Finally, the chapter will discuss the process and outcome of acceptability testing of the intervention materials.

8.1.1. Intervention content: theoretical background

8.1.1.1. Acceptance and Commitment Therapy

Psychological flexibility and self-compassion were identified as potentially protective variables that mitigate psychological distress in parents of children with a visible difference. Psychological flexibility and self-compassion are central processes (Harris, 2019) within the therapeutic model of Acceptance and Commitment Therapy (ACT). With the aim of promoting psychological wellbeing, ACT is a “third generation” transdiagnostic behavioural therapy which encourages individuals to build an awareness of their personal values and commit to living in accordance with them (Hayes et al., 2006b) by teaching individuals skills to help manage difficult thoughts and feelings that may prevent them from taking valued action (Dindo et al., 2017). Unlike Cognitive Behavioural Therapy (CBT), ACT does not focus on altering or challenging thoughts, but instead encourages individuals to practice acceptance of difficult internal experiences and use strategies to continue to live in accordance with personal values (Luoma et al., 2007).

ACT theory is based on the premise that the central mechanisms which cause psychological suffering are experiential avoidance (attempting to prevent or alter cognitions, feelings, or physical sensations) and cognitive fusion (identifying closely or becoming attached to and acting in line with difficult thoughts) (Hayes et al., 2006b). Through these processes, behaviour is controlled by one's thoughts, feelings, and other internal experiences, rather than personal values. This process has been labelled psychological **in**flexibility. ACT also acknowledges that suffering is a natural and unavoidable part of being human but that individuals do not necessarily need to resign themselves to suffering. Therefore, ACT is focussed on cultivating psychological flexibility. Psychological flexibility is the ability "to contact the present moment" with awareness and "change or persist in behaviour" in accordance with values (Hayes et al., 2006, p.7). Psychological flexibility is therefore a key mechanism in reducing the negative impact of experiential avoidance, which steers individuals away from value-based action. ACT uses metaphors as a technique to communicate the concepts and processes of psychological flexibility in accessible and memorable language and format (Luoma et al., 2007).

When compared to other parent intervention approaches, the ACT-based approach provides an opportunity to focus on parents own emotional, cognitive, and behavioural responses to their child's condition or injury. As described previously (section 2.6.7.), much of the existing support for parents in this field is focussed on practical skills training, rather than supporting parents and carers with their own psychosocial challenges. For example, the Triple P

Positive Parenting Program delivered to parents of children with eczema focussed on parenting skills related to child behaviour, illness management, and parenting practices (Morawska et al., 2016). As such, the psychosocial needs of parents themselves are not the focus of such interventions. The findings from study 1 and 2 demonstrate that parents of children with visible differences may experience psychological distress and stress. Consequently, the present intervention was constructed using a therapeutic approach that could equip parents with the skills to manage difficult thoughts and feelings and thus address this gap in support provision. The next section of this chapter will explore the core processes of the ACT approach in greater detail.

8.1.1.2. The ACT model: six core processes of ACT

ACT targets six core processes with the overall aim of increasing psychological flexibility (Hayes et al., 2006).

Acceptance

Acceptance or experiential acceptance is presented an as alternative to experiential avoidance (Hayes et al., 2006). The process of acceptance encourages individuals to ‘open up’ and ‘make room’ for unwanted or difficult private experiences (e.g., thoughts and feelings). Rather than struggling with or resisting challenging thoughts and feelings, the acceptance process teaches individuals to allow themselves to experience them without judgement.

Cognitive defusion

ACT attempts to alter the way an individual interacts with and relates to their thoughts (Hayes et al., 2006). Cognitive defusion encourages individuals to take a ‘step back’ or detach from thoughts, images, and memories (Harris, 2019).

Being present

ACT encourages ongoing contact with the present moment (Hayes et al., 2006). This process is taught through exercises that promote the ability to consciously and flexibly pay attention to what is happening around you, as it happens (Harris, 2019). This can include attending to both the physical space around us and the psychological experiences occurring within us (e.g., cognitions and emotions). Being in contact with the present moment can enable individuals to improve their understanding of how they feel, think, and react, as well as being able to connect deeply with other people (Harris, 2019).

Self-as-context

This process examines the way in which individuals relate to themselves, versus the way they perceive and relate to other people (Barnes-Holmes & Roche, 2001). ACT distinguishes between the 'thinking mind' (the part that is generating thoughts, beliefs, recalling memories etc.) and the 'noticing mind' (the part that is aware of what we are doing, feeling, and thinking, Harris, 2019). Self-as-context is the process concerned with the 'noticing mind' and its ability to observe cognitions, emotions, and behaviours. Similar to defusion, this process allows individuals some distance between themselves and their thoughts and feelings.

Values

Values are chosen desired qualities of physical or psychological action (e.g., honesty, courage, compassion; Harris, 2019; Hayes et al., 2006). They are not a goal or object that can be obtained, but instead describe how an individual might like to behave on a long-term basis. There are several exercises in ACT that are designed to help individuals identify and clarify their values (Hayes et al., 2006). Identifying values can help to cultivate motivation for action and guide goal setting (Harris, 2019).

Committed action

ACT promotes the development of action and behaviours which are in accordance with an individual’s values (Harris, 2019; Hayes et al., 2006b). Committed action includes physical action (the way we choose to behave) and psychological action (our internal thoughts and feelings; Harris, 2019). ACT theory acknowledges that taking value-based committed action might result in an individual experiencing difficult thoughts and feelings (Harris, 2019). Therefore, committed action requires processes similar to other behavioural therapies such as problem solving, goal setting, behavioural activation and exposure, and skills to manage challenging thoughts and emotions, such as mindfulness and self-soothing (Harris, 2019; Hayes et al., 2006).

The six core processes can be grouped into two overarching themes that are present throughout ACT. Acceptance, cognitive defusion, being present and self-as-context can be categorised more broadly as “mindfulness and acceptance processes”, whereas values, committed action, being present, and self-as-context are all related to “commitment and behaviour change processes” (Hayes et al., 2006). See Figure 20 for a visual representation of the groupings of these core processes.

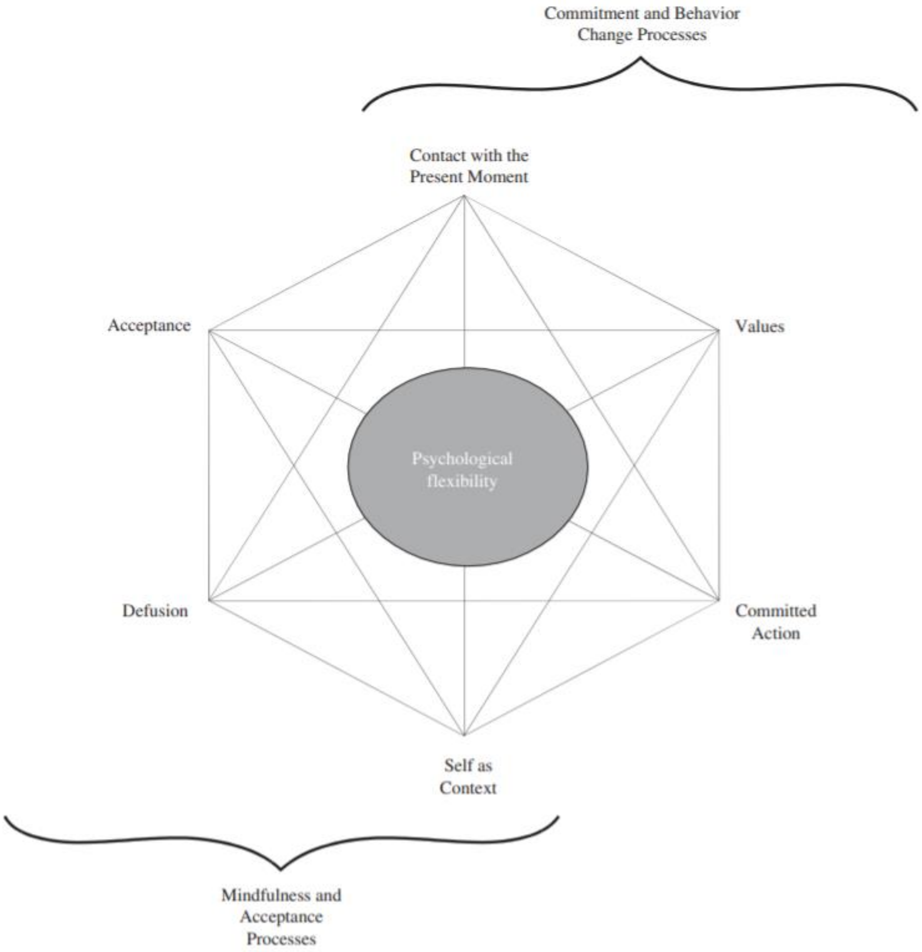


Figure 20: Diagram of ACT model illustrating the six core processes.

Self-compassion and ACT

The mixed methods integration process (see Chapter 7) found that self-compassion was important in the process of helping parents manage their thoughts and feelings related to their child's visible difference. Specifically, the quantitative data found that self-compassion mediated the relationship between parent-reported psychological flexibility and stress difficulty. This suggests that self-compassion has a vital role in the association between psychological flexibility and psychological wellbeing.

Russ Harris, a leading expert in the field of ACT, argues that self-compassion is a value that is embedded within every aspect of the ACT model (Harris, 2019). Harris (2019) posits that we can define self-compassion simply as “acknowledging pain and responding with kindness to oneself” (Harris, 2019, p. 265). Self-compassion is closely linked to acceptance, a core principle of ACT. Acceptance requires individuals to acknowledge and allow room for experiences, which requires kindness and self-compassion (Harris, 2019). Alongside the theoretical links within the ACT model, empirical evidence has highlighted the association between self-compassion and psychological flexibility. A survey of 114 university students found that self-compassion was significantly correlated with psychological flexibility processes (Marshall & Brockman, 2016). In addition, self-compassion predicted significant unique variance beyond psychological flexibility across various psychological wellbeing outcomes. Consequently, although acceptance is theoretically linked to compassion, it was important to ensure that self-compassion was an explicit and integral to intervention development and interwoven throughout content and activities.

8.1.1.3. ACT interventions for parents and carers

ACT is an approach that has been utilised in interventions for parents and carers of children with paediatric chronic illness. An acceptability and feasibility study of an online ACT intervention (iACT-P workshop) for parents of children with Inflammatory Bowel Disease found that retention rate across the three sessions was high (>70%, Kohut et al., 2021), and parents who attended the intervention also provided positive qualitative feedback (e.g., feeling validated in their experiences). An ACT intervention, known as “Take a Breath”, has also been developed for parents of children with cerebral palsy. Comprised of six group sessions and facilitated using a video conferencing format (McMillan et al., 2020), initial acceptability and feasibility testing of this intervention found that parents experienced significant reductions in guilt, worry, and increased mindfulness. Finally, in a pilot eight-week group-based intervention (unnamed) with 8 parents of children with chronic pain, an increase in pain-related psychological flexibility was found in parents who completed the intervention (Wallace et al., 2016). Parents also indicated high satisfaction with the intervention.

Although, preliminary evidence suggests that ACT could be a beneficial therapeutic model for support for parents of children with long-term health conditions, the interventions discussed above are group based and facilitated by professionals, rather than being self-administered. Therefore, these findings may not be completely generalisable when considering the development of a self-administered ACT intervention for parents.

8.1.1.4. ACT in a visible difference population

Zucchelli et al. (2018) have provided a sound theoretical argument for the use of ACT with this population, particularly in relation to its focus on experiential acceptance. Unlike other behavioural therapies, such as CBT, ACT does not require individuals to challenge or alter their cognitions. Instead, it focusses on the process of acceptance and the ability to detach from difficult thoughts and feelings, to live a meaningful life. For individuals with a visible difference, upsetting thoughts and feelings may actually reflect their reality (Zucchelli et al., 2018). For example, an individual with a visible difference may experience negative thoughts about their appearance (e.g., *my appearance is not normal*) or anxieties that they may receive unwanted attention (e.g., staring, abusive comments) that are difficult to dispute or are outside of their control, thus experiential acceptance and the ability to defuse may be more appropriate than trying to challenge thoughts and feelings. ACT also does not frame these kinds of cognitions as maladaptive or dysfunctional, but instead focuses on the possible impact of thoughts on value-based action, through cognitive fusion or experiential avoidance. As a result, ACT does not target thoughts directly, individuals are not required to alter their cognitions or self-evaluations related to their appearance (Zucchelli et al., 2018), rather ACT encourages individuals to develop psychological flexibility and hold difficult thoughts and feelings at a distance, with the aim of promoting value-based action.

Empirical evidence in the field of visible difference has also identified the role of psychological flexibility in promoting wellbeing. A cross-sectional survey study of 120 women with lipoedema (progressive visible enlargement of legs due to swelling and deposition of fat), found that self-reported psychological flexibility significantly positively predicted quality of life (Dudek et al., 2016). A cross-sectional survey study of 78 burn injured patients also found that increased appearance-related anxiety was significantly related to reduced acceptance, cognitive defusion and committed action (Shepherd et al., 2019). A cross-sectional survey study with 220 adults with a range of visible difference found that cognitive fusion partially mediated the relationship between body evaluation and experiential avoidance, as well as the relationship between body evaluation and appearance-fixing behaviours. Additionally, it was found that experiential avoidance mediated the relationship between body evaluation and behavioural avoidance (Zucchelli et al., 2020). These findings suggest that engagement in processes that promote psychological flexibility can be protective against psychological distress. The evidence also suggests that psychological flexibility may be beneficial in the development of adaptive strategies for coping with negative body evaluation and appearance-related distress. However, it is important to note that all the literature discussed utilised a cross-sectional design thus it is not possible to draw conclusions about the longitudinal impact of psychological flexibility on psychological wellbeing in this population.

The existing literature exploring the benefit of ACT for parents of children with a visible difference is scarce. Although the condition-specific research has found self-compassion to be protective against negative affect (Hawkins et al., 2019), the present PhD is the first to explore the role of psychological flexibility in parents of children with a range of appearance-affecting conditions and injuries. Therefore, the development of the present intervention has taken an entirely novel approach to support for this population.

Although there are no interventions based on ACT for parents of children with a visible difference, a psychosocial intervention based on the principles of ACT have been developed for individuals with a visible difference. ACT It Out is a mobile health app developed to support adults with a visible difference experiencing common psychosocial challenges (Zucchelli et al., 2021). Initial stakeholder feedback on ACT It Out indicated that user representatives and clinicians felt that ACT-based content delivered via digital format was relevant and beneficial for adults with a visible difference (Zucchelli et al., 2021).

Based on the integrated mixed methods findings, the existing literature, and intervention development in the field of visible difference, ACT was deemed an appropriate therapeutic model on which to base an intervention for parents of children with a visible difference.

8.1.1.5. Family coping

There is a wide array of theory and literature which explores family coping. Within these theoretical models, the psychosocial constructs of flexibility, communication, and problem solving have all been identified as important to family functioning (Miller et al., 2000; Olson, 2000). The Circumplex Model of Family Systems (Olson, 2000, see section 6.4.3. and 6.4.4.) describes a flexible family as one with shared leadership, role-sharing, and change occurring when appropriate. These authors hypothesised that balanced flexibility (structured but flexible) is beneficial for family functioning. This is not dissimilar to the role that psychological flexibility plays for an individual, wherein an individual does not become enmeshed with thoughts and feelings and is open to change.

Communication has been highlighted as being important in several models of family functioning (e.g., Miller et al., 2000; Olson, 2000). The Circumplex model of Family Systems states that effective family communication includes a variety of speaking and listening skills. This includes speaking with clarity, self-disclosure, respect, empathy, and attentive listening. The McMaster family model defines communication as “how information is exchanged within the family” and is subdivided into instrumental communication (practical issues that occur in everyday life) and affective communication (Miller et al., 2000). Both models indicate that open and effective communication is an important contributing factor in the development and maintenance of adaptive family functioning. However, both models also focus mostly on verbal communication, and are therefore limited in their ability to account for the role of non-verbal communication in families.

Multiple models of family functioning also include the ability to problem solve. Problem solving within families has been defined as “a family’s ability to resolve problems at a level that maintains effective family functioning” (Miller et al., 2000, p.170). Problem solving for parents of children with a visible difference could reflect a variety of challenges, such as treatment decision making, responding to social challenges, attending appointment etc. The Circumplex Model of Family Systems identifies aspects of effective communication which are also important in problem solving in couples and families (Olson, 2000). Therefore, some elements of managing challenges related to a child’s visible difference may be interconnected with developing and maintaining good communication skills within the family unit. However, this model does not examine how communication of information from external sources (e.g., information from health care providers) may also be important to family problem solving.

Existing models lend support to the inclusion of content related to promoting flexibility and open and effective communication in an intervention for parents of children with a visible difference. This chapter will now consider the process of intervention development.

8.1.2. Intervention development

This section will discuss the process of intervention development. The stages of intervention development, as outlined in the MRC guidelines, will be considered. The collaborative nature of the Participatory Action Research (PAR) approach to intervention development is discussed. Additionally, the development of the content and format will be described. A copy of the intervention materials following iterative development can be seen in Appendix M.

8.1.2.1. MRC guidelines for development of complex interventions

The updated MRC guidelines (Skivington et al., 2021) consider a broader conceptualisation of complexity than the previous 2008 guidelines (see section 3.12. for greater detail). The new framework has retained the main stages: 1) development, 2) feasibility, 3) evaluation, and 4) implementation, but unlike the previous guidelines it requires researchers to consider a number of core elements at each stage of the intervention research (See Figure 12). The guidelines state that the development of a new intervention should be based on evidence and theory (Skivington et al., 2021). Study 1 and 2 were focussed on exploring the experiences and support needs of parents of children with a visible difference. Following the integration of the findings of these two studies, overarching psychosocial constructs were identified and utilised to develop a draft of the intervention materials. As shown in Figure 21, several core elements were considered during this development stage.

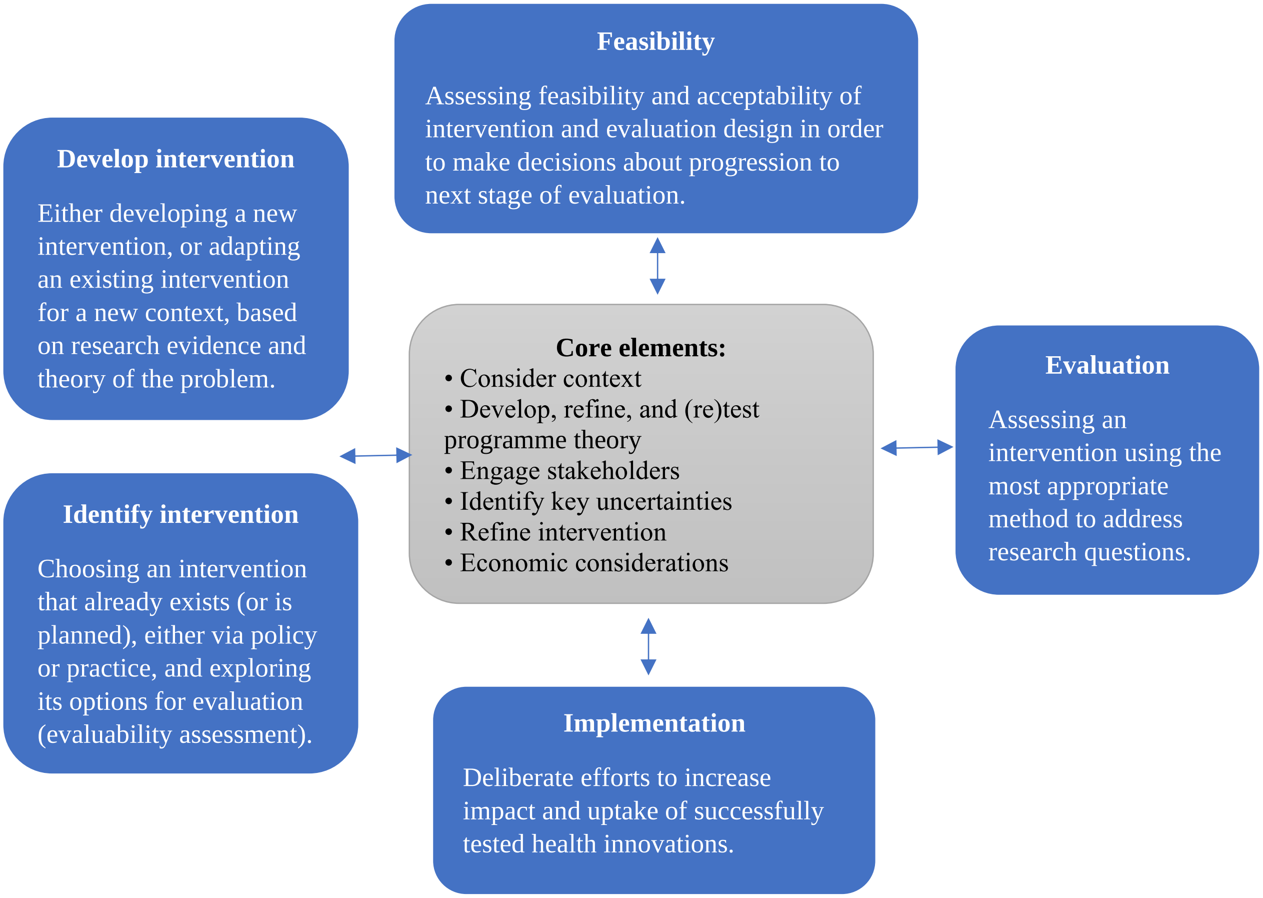


Figure 21: Framework for developing complex interventions (Skivington et al., 2021)

The development of programme theory (key components of the intervention, how they interact, and the mechanisms of the intervention) were the primary focus of the development phase (Skivington et al., 2021). The present PhD followed best practice by developing theory alongside stakeholder involvement, drawing on existing evidence and theory from relevant fields. This theory was added to and refined as the PhD mapped qualitative findings onto the quantitative design, and again during the mixed methods integration of the findings. This chapter builds upon the previous development work by utilising a Participatory Action Research approach (see section 8.1.3.). Engagement with public involvement representatives and other key stakeholders provided an opportunity to gain feedback on the proposed content and mode of delivery. In response to this feedback, the intervention was iteratively refined and changed whilst considering all the core elements of the MRC framework (Skivington et al., 2021). The Participatory Action Research approach and its application within this intervention development process will be described in section 8.1.3.

8.1.2.2. Intervention materials: developing content

All intervention content was based on the integrated mixed methods findings and existing literature from the visible difference field (e.g., Heath et al., 2018; **Thornton** et al., 2021; Zelihić et al., 2021; Zucchelli et al., 2018, 2020). Some aspects of the intervention were informed by existing advice or support on specific topics (e.g., bullying, communication etc.). Several of the metaphors used in the ACT-based content (e.g., passengers on a bus) were informed by materials developed by Russ Harris that have been shared in the public domain (Harris, 2019). Exercises were adapted from the ACT Now training materials (designed for equipping health professionals with the skills to deliver ACT-based support to individuals with appearance-affecting conditions and injuries (e.g., breathing exercise), which have been found to be acceptable (Zucchelli, in press). The Explain, Reassure, Distract exercise for managing the reactions of others was also adapted from resources shared by Changing Faces (Explain, Reassure, Distract exercise, Changing Faces, 2020). Furthermore, the story telling exercise was informed by existing literature with parents of children with childhood cancer. In a study with the families of 28 children with cancer, a digital storytelling intervention was found to have high feasibility and parents reported that the intervention provided emotional comfort, facilitated parent-child communication, and was an effective coping strategy (Akard et al., 2015). Therefore, a story telling exercise using a similar format was designed for the present parenting intervention. Finally, the intervention sections on bullying and cyber bullying were informed by resources from an online platform specialising in advice and guidance on bullying in children and young people (Bullying UK, 2021).

All intervention content was drafted by the PhD student and iteratively reviewed and discussed with the supervisory team at multiple points throughout the process. Following each feedback point, the PhD student made changes to the intervention materials before the next stage of feedback. The supervision team included a qualified Clinical Psychologist with experience of working clinically with individuals with a visible difference, and their parents. See Figure 22 for a flowchart illustrating the iterative intervention development process. The PhD student also worked with a professional illustrator to create bespoke images to reflect and enhance the textual content. The textual intervention content was assessed to be presented at a reading age of 12-years-old or 8th grade.

The intervention materials were divided into two main sections. The first section entitled "core skills for parents" focuses on five of the six core principles of ACT (acceptance, cognitive defusion, being present, values, and committed action) and taught skills for managing difficult thoughts and feelings, known as "helper skills". Helper skills were based on the two overarching concepts of ACT: being present and values-based action. See Appendix J for a full breakdown of how the intervention sections map onto the core principles of ACT. The concept of self-as-context was not included explicitly in the intervention materials. Experts in the field state that self-as-context is the most complex concept within ACT and can be difficult for service users to understand and apply independently (Harris, 2019). However, self-as-context is implicit within other processes and related exercises (e.g., being present and mindfulness exercises). Due to the self-administered nature of the materials, it was decided not to include explicit mentions of self-as-context to maintain accessibility of the content. The second section of the intervention was comprised of advice, guidance, and exercises on the key constructs and topics identified in the mixed methods integration: managing social challenges (e.g., the reactions of others), communication, finding information, and finding the right support. The exercises throughout the intervention provide interactive options for users to make notes, record thoughts and feelings, as well as values and goals.

Health research has found that psychoeducation is often not sufficient to change behaviour and there can be significant unique benefit from combining psychoeducation and therapeutic elements (Dowd et al., 2015; Zaretsky et al., 2008). For example, a comparison of psychoeducation programs and mindfulness-based cognitive therapy (MCBT) for pain management found that although both groups demonstrated improvements in subject well-being, these were more pronounced in the MCBT group (Zaretsky et al., 2008). In addition, participants in the MCBT group reported greater reduction in pain and increased ability to manage emotions, stress, and enjoy pleasant events. This suggests therapeutic elements of an intervention can be more beneficial than psychoeducation alone. Therefore, the intervention materials combine psychoeducational content and interactive ACT-based activities.

8.1.2.3. Intervention delivery and intensity

Once the key content had been identified, it was vital to consider the level of intervention and mode of delivery. The existing literature has found that the self-directed format of the Triple P Parenting intervention led to significant improvements of both parent and child outcomes (Sanders et al., 2014). A systematic review of parenting interventions found no significant differences in parent-reported measures of child outcomes when comparing self-directed interventions with and without therapist input (Sanders et al., 2014). This review also found that self-directed parenting interventions significantly improved parental mood and parenting efficacy and reduced parenting stress. This effect was maintained even once self-directed interventions with therapist input were removed from the analysis (Sanders et al., 2014). The existing literature has also stated that evidence-based self-help parenting interventions may be a cost-effective method of increasing access to psychosocial support (Daley & Rydon-Grange, 2013). Previous PPI input (see section 3.11.) indicated that a psychoeducational workbook of resources on different relevant topics would be beneficial for this population. Consequently, the intervention materials are presented in a self-administered online workbook format entitled “The Parenting Toolkit”. The intervention materials utilised a combination of text, professionally designed bespoke images, and video and audio links.

The mixed methods findings demonstrated that most parents experienced some level of psychological distress in relation to their child’s visible difference. However, many of these individuals reported that this distress did not persist at a high intensity over a long period of time. The majority of this population require low intensity support, but given the widespread experience of this distress, this support also needs to be easily accessible to a large number of people. Psychological support for this population is scarce, therefore developing a low intensity intervention could reduce demand on limited resources (e.g., trained and available practitioners) and provide support that is acceptable to those with less complex and low risk needs.

The Centre for Appearance Research (CAR) framework for appearance-related interventions (Figure 22, Rumsey & Harcourt, 2012) was developed to identify the most appropriate methods for implementation and evaluation of interventions for individuals affected by a visible difference. The CAR framework categorises levels of psychosocial interventions using a stepped care model. The levels of intervention range from level 0 (general population campaigns) to level 5 (complex, specialist led counselling/therapy). The shape of the framework is designed to indicate the shifting intensity of intervention (0 = low, 5 = high) and the number of individuals who require that level of intervention (0 = many, 5 = few). Based on the mixed methods findings and existing literature, the present intervention was developed to be self-administered, independent from other interventions and not reliant on input from health or support professionals. Due to this mode of delivery and the low intensity therapeutic content, this intervention is most closely aligned with level 2 (stand-alone intervention) of the CAR framework.

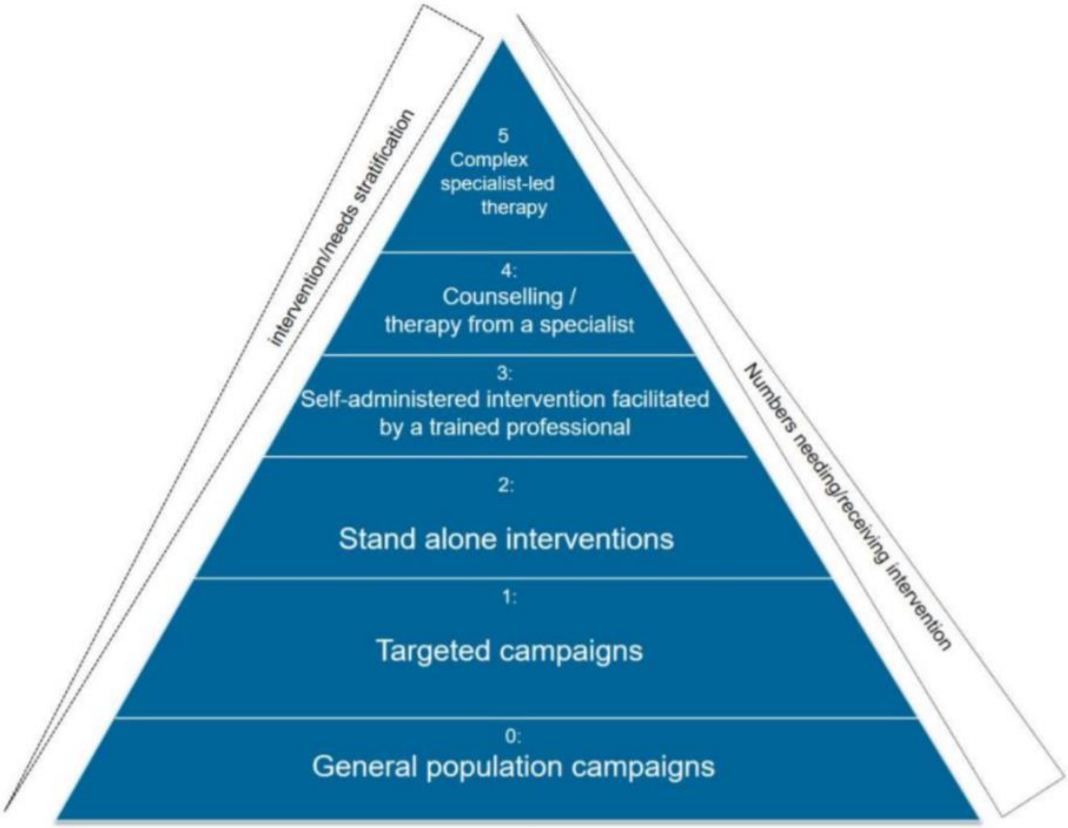


Figure 22: The Centre for Appearance Research framework of appearance-related interventions (Rumsey and Harcourt, 2012)

8.1.3. Participatory Involvement

The present study employed a Participatory Action Research (PAR) approach. PAR is a collective and self-reflective approach to inquiry which involves members of the public who have lived experience of the topic of the research (Baum et al., 2006). Public involvement representatives should include individuals who have experiences and hold perspectives which are important to the research question (National Institute for Health Research, 2022). For the present research it was important to attempt to include a range of parent perspectives from different backgrounds (e.g., parents of children of different ages, different visible differences, different genders etc.).

PAR is a collaborative research approach rather than a discrete methodology (Turnbull et al., 1998), in which researchers and representatives engage in a reflective practice to design and conduct research that aims to enable change and action (Baum et al., 2006). The PAR approach advocates for power in research to be shared between the researcher and representatives (Baum et al., 2006). In this way, the researchers and lived experience representatives become partners in the research process. PAR challenges the systems of knowledge control held by traditional research communities and encourages members of the public to become actively engaged and influence the direction of research (Baum et al., 2006).

Parents in family research can have important benefits for both researchers and families (Shen et al., 2017). Parents and caregivers are usually the constant in a child's life and can bring a great deal of expertise about the child's health experience to the research. This ensures that research is more likely to be meaningful and sensitive to the needs of children and families (Shen et al., 2017). Involvement in research has been found to elicit a sense of empowerment in parents, as well as increased confidence and the opportunity to learn new skills (Shen et al., 2017).

Health and support professionals who work with families of affected children were also asked to review and provide feedback on the intervention. Health professionals are often the first point of contact for these families and have insight into concerns and challenges faced by this population (Gee et al., 2020b), and so are well placed to give feedback on the content of the proposed intervention.

The use of a PAR approach can be particularly important in intervention development. It is critical that stakeholders and users are involved in the early stages of intervention development to ensure it reflects future users' experiences and support needs and maximise its relatability, as acceptability can affect implementation, effectiveness, and motivation (Nastasi et al., 2000). When PAR has been used as a model for intervention development with individuals with visible differences, the resulting psychosocial interventions have been found to be highly acceptable (e.g., YP Face IT, Williamson et al., 2015).

8.1.3.1. Application of Participatory Action Research approach

Parents who had participated in previous public involvement activities were contacted via email to ask if they were interested in contributing to the intervention development. Charitable organisations who had previously supported recruitment (e.g., Appearance Collective charities) were contacted via email with information about the project and asked if any parent members would like to contribute to the intervention development. Due to the cross-condition nature of the present PhD, a purposeful sample of parent public involvement representatives with children with a range of conditions and injuries was recruited. Health and support professionals with clinical experience of working with parents of children with a variety of appearance-affecting conditions and/or injuries and who had previously expressed an interest in the PhD research were contacted via email and asked if they would be interested in providing feedback on the development of the intervention.

Five parents of children with a range of appearance-affecting conditions and three health and support professionals took part in the public involvement workshops. All parents identified as female, and the child’s mother. Two of the children of these parents were female and three were male. All health and support professionals also identified as female. More demographic information about the members of the public involvement group can be found in Appendix K.

Parent user representatives and health and support professionals were invited to attend separate group workshops facilitated using Microsoft Teams video conferencing software. They were provided with information about the research project and the proposed outline for the content of the intervention to review prior to the workshop. One of barriers identified to engaging in PPI is lack of clarity around roles and input (Froggatt et al., 2015). Therefore, parent public involvement representatives (parents with lived experience of caring for a child with an appearance-affecting condition/injury) were provided with a session of PPI training prior to the public involvement workshops. The discussion guides for the public involvement workshops can be found in Appendix K.

Workshop sessions were recorded and transcribed. Using template analysis, four primary themes were identified within the feedback given by the parents and professionals during the workshops: 1) Meeting support needs, 2) Communicating complex concepts, 3) “Getting parents on board”, and 4) Accessibility of format. The full template analysis with exemplar quotes can be found in Appendix K. Feedback (not always consistent) recommending changes to the intervention was collated into a table of changes (see Appendix K). Balancing conflicting feedback from multiple sources (e.g., parents versus health professionals) requires researchers to balance and prioritise evidence, personal experience, and competing values held by each public involvement member or group (Hoddinott et al., 2018). When conflicting feedback arose, the experience and values within the group was considered on a case-by-case basis, and in light of past relevant theory and evidence, to determine alterations to the intervention.

The first theme, “Meeting support needs” described whether the intervention materials met parent support needs. Both parent and professional representatives identified areas in the support materials where they felt the needs of parents had been well represented in the content (e.g., skills for parents to manage their own thoughts and feelings) and many commented generally that the materials addressed a gap in parent support. Parents valued the flexibility of the materials. This included the ability to dip in and out as needed throughout the child’s life and the choice of approaches and activities that could be adapted to meet the needs of different situations and families. Professionals also reported that they could see themselves using the intervention materials during therapeutic sessions with families. However, some identified areas where content could be enhanced, or additional information included (e.g., advocacy for children within healthcare settings, the role of school and educators).

“Communicating complex concepts” describes feedback about the challenge of breaking down complex therapeutic concepts (e.g., defusion, values) into an accessible and clear narrative that parents from a range of backgrounds could follow independently. Many agreed that the content of the intervention materials was presented in an accessible way and commented on the success of various tools (e.g., metaphors, examples of typical concerns) used to break down complex or abstract concepts. In particular, parents felt the content validated their own concerns and would help users appreciate that others experience similar challenges. Some parents expressed difficulties with comprehension of some concepts (e.g., passengers on bus metaphor and evolutionary explanation of difficult thoughts and feelings). To address these concerns, the section of evolution was reduced and the language within the passengers on a bus metaphor was edited to increase clarity, in line with PAR feedback.

Parent engagement in the intervention materials was discussed throughout the workshops. “Getting parents on board” reflects feedback around the challenges of encouraging caregivers to focus on their own needs, rather than just the needs of their child. Both parents and professionals emphasised the necessity to build trust with intervention users, especially given that the materials may touch on sensitive or emotive topics. Parents and professionals expressed that the introduction section outlined the benefits of seeking support, for both parent and children. In addition, the title “The Parenting Toolkit” and the tag line “Supporting your child with an appearance-affecting condition or injury” clearly communicated the aim and target audience of the materials. Although parents felt that the introductory section flowed well, in line with recommendations from the professional representatives, a summary of content of the materials was added to encourage parents to continue reading.

All representatives contributed to discussion about the layout and format of the intervention materials in some capacity. The use of language was discussed in the context of whether certain terms common in therapeutic settings would be accessible in a self-administered format. The length and presentation of the content was also discussed, with parents and professionals providing suggestions for additions and changes (e.g., slight alterations in language, increased signposting to information using contents page and text layout).

The steps of the iterative development process are illustrated in Figure 23. Overall, the feedback from parents and professionals was positive and suggested that the present intervention would meet the needs of this population. Feasibility and acceptability testing is the next phase of intervention development in accordance with the Medical Research Council (MRC) framework (Skivington et al., 2021).

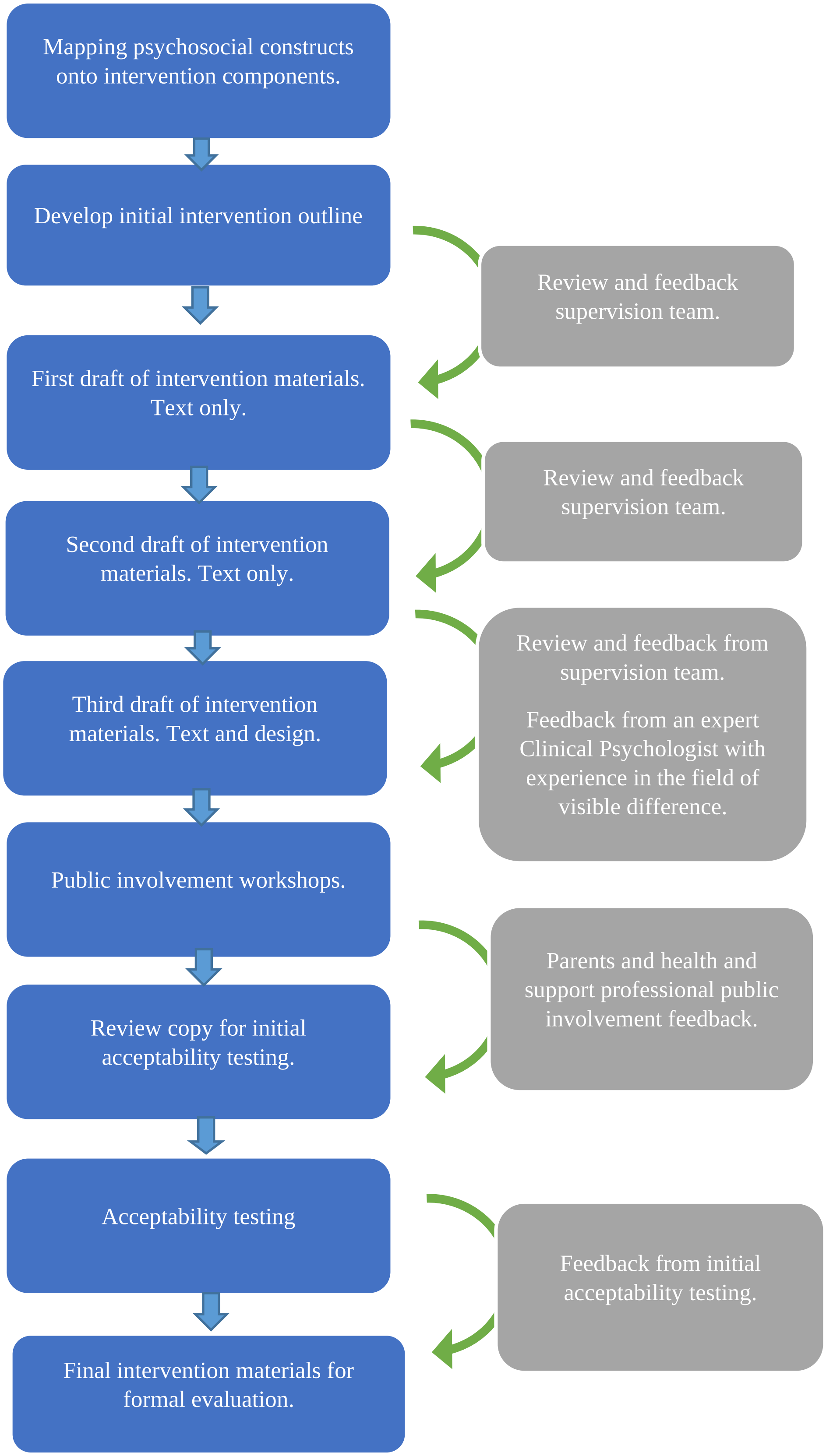


Figure 23: Diagram of integrative intervention development process

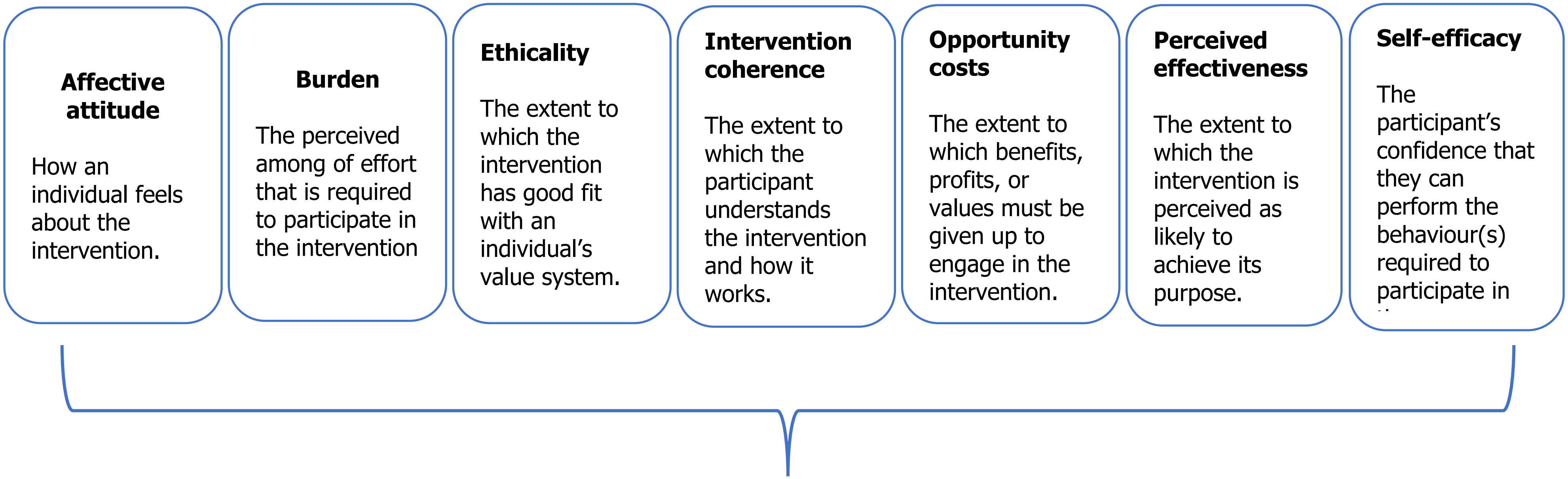
8.1.4. Feasibility and acceptability

Feasibility testing is the second phase of intervention development outlined in the MRC framework. It has been proposed that feasibility research should be designed to assess predefined criteria that relate to either the evaluation design or the intervention itself (Skivington et al., 2021). When the intervention is the focus of feasibility research, there are several aspects that could be considered including content, delivery, acceptability, adherence, cost-effectiveness or capacity of providers (Skivington et al., 2021). Consideration of these factors provides opportunity for iterative change and refinement of the intervention materials. The present study focussed on exploring and understanding the acceptability of the content and delivery of the intervention.

It has been established that assessment of acceptability is a critical element of implementation and thus overall effectiveness of interventions (Nastasi et al., 2000). It is important that the content and delivery of interventions be acceptable to both intervention deliverers and recipients (Diepeveen et al., 2013; Stok et al., 2016). If interventions have low acceptability for either of these groups, the intervention will not be delivered or received as intended (Sekhon et al., 2017). This will also influence overall effectiveness of the intervention. In the present intervention, parents of children with a visible difference occupy the roles of both the deliverer and the recipient, therefore gaining acceptability data from this population is essential. However, definitions of acceptability vary significantly (Sekhon et al., 2017). This poses a significant challenge when attempting to assess or measure this construct in intervention development. The following section will describe a systematically developed framework of acceptability for healthcare interventions, designed with the aim of providing a clear, shared understanding of the concept.

8.1.5. Theoretical Framework of Acceptability

Acceptability in research has been defined as “a multi-faceted construct that reflects the extent to which people delivering or receiving a healthcare intervention consider it to be appropriate, based on anticipated or experiential cognitive and emotional responses to the intervention” (Sekhon et al., 2017, p.4). The Theoretical Framework of Acceptability (TFA) was constructed following a rigorous process of reviewing existing definitions of acceptability (Sekhon et al., 2017). Firstly, the authors conducted a review of 43 systematic reviews which have previously aimed to define, theorise, or measure the acceptability of healthcare interventions. Following this review, inductive and deductive processes were utilised to develop a theoretical framework of acceptability. The process of developing this framework included the following steps: 1) defining acceptability, 2) describing its properties and scope, and 3) identifying component constructs and empirical indicators. The TFA outlines seven key components of acceptability (Sekhon et al., 2017). See Figure 24 for the components of TFA.



Affective attitude

How an individual feels about the intervention.

Burden

The perceived among of effort that is required to participate in the intervention

Ethicality

The extent to which the intervention has good fit with an individual's value system.

Intervention coherence

The extent to which the participant understands the intervention and how it works.

Opportunity costs

The extent to which benefits, profits, or values must be given up to engage in the intervention.

Perceived effectiveness

The extent to which the intervention is perceived as likely to achieve its purpose.

Self-efficacy

The participant's confidence that they can perform the behaviour(s) required to participate in

Prospective acceptability: prior to engaging in the intervention

Concurrent acceptability: whilst participating in the intervention

Retrospective acceptability: after participating in the intervention

Figure 24: The Theoretical Framework of Acceptability (TFA), Sekhon et al., 2017

As shown in the Figure 24, the TFA also distinguishes between assessing acceptability at different time points in relation to their interaction with the intervention. The inclusion of this temporal element was to reflect the measurement of acceptability before, during, and after intervention delivery as found in the systematic review process (Sekhon et al., 2017). This is important to reflect upon when considering the present intervention, as the temporality of acceptability testing can influence the design and purpose of the assessment or evaluation. The present study took a retrospective approach (Sekhon et al., 2017) because participants were required to reflect on the intervention after they had an opportunity to review and engage with the content.

The TFA was developed after considering reviews of acceptability studies of complex healthcare interventions (Sekhon et al., 2017). TFA draws on health psychology theory, specifically illness perceptions literature (Moss-Morris et al., 2002), to conceptualise facets of acceptability missing from previous definitions (e.g., intervention coherence) and therefore enhance the final framework. Due to links to health psychology literature and theory, the TFA was considered an appropriate framework to consult during acceptability testing.

The following chapter will address the feasibility and acceptability testing of the parenting intervention, developed based on the mixed methods findings. The aim of the study was as follows:

Aim:

1) To assess the feasibility and acceptability of a self-administered ACT-based intervention for parents of children with range of visible differences.

Objectives:

- Identify whether the content and topics covered in the intervention meet the support needs and reflect the experiences of a broader sample of parents of children with a visible difference
- To assess whether the content and format of the materials is accessible to a broader sample of parent users

8.2. Method

8.2.1. Study design

Data were collected using an online survey comprised of self-report measures. Compared with an interview design, an online survey facilitated participation from a large and more heterogenous sample of parents and carers (e.g., range of conditions and injuries, caregiver roles, family structure etc.). Similar to the online survey design of study 2 (see section 6.2.7.), qualitative data were also collected to enhance and clarify the quantitative data and provide participants an opportunity to elaborate upon their responses in their own words (Greene et al., 1989). Therefore, the qualitative data were used for complementary purposes and were integrated with the quantitative data using a ‘following a thread’ approach (See section 6.2.7.6., Moran-Ellis et al., 2006).

8.2.2. Survey design

The survey (see Appendix L) was designed based on the existing visible difference literature (e.g., Heath et al., 2019), acceptability testing theory (Sekhon et al., 2017), and previous findings in the PhD. Items from an existing acceptability scale were examined alongside the TFA, to assess whether the instrument addressed the components present in the framework. The outcome of the PAR workshops were also considered during the survey design. This was to ensure that the themes raised in the feedback workshops were also represented in the survey. All materials were reviewed by parent public involvement representatives.

eHealth Impact Questionnaire

The survey included two subscales adapted from part two of eHealth Impact Questionnaire (eHiQ; Kelly et al., 2015):: the confidence and identification subscale and the presentation of information subscale. These subscales were selected because they best represented the constructs outlined within the TFA (see section 8.1.5., Sekhon et al., 2017). In addition, this scale has previously been utilised in a study assessing the acceptability of a website for parents of children with burn injuries (Heath et al., 2019), which suggests this scale is appropriate for the present parent population. The eHIQ was initially designed for assessing acceptability of informational health-focussed websites. Therefore, the wording of the items was altered to refer to the "parenting toolkit". The original items were also designed for individuals seeking information and advice for their own health. These items were adapted to be suitable for parents seeking support for themselves, whilst also referring to their child's condition rather than their own.

The third subscale of the eHIQ was not included. The third subscale was focussed on understanding and motivation and the items were closely related to outcomes of information seeking (e.g., feeling informed to make decisions) and individual coping. The parenting toolkit has a cross-condition therapeutic focus, it is not a tool for delivering information about condition-specific challenges. Additionally, public involvement representatives who had viewed the intervention materials also commented that the items in this subscale were less relevant. This subscale did not meet the aims of this study and was not included.

The confidence and identification subscale included 9 items rated on a five-point Likert scale from strongly disagree (1) to strongly agree (5). Some example items from this subscale include: "The people who have contributed to the parenting toolkit understand what is important to me and my family" and "The parenting toolkit gives me more confidence to support my child". As well as mapping onto constructs of the TFA (e.g., self-efficacy, ethicality, perceived effectiveness) this subscale reflected the issues discussed in several themes identified in the PAR workshops (e.g., “Getting parents on board” and “Meeting support needs”).

The information and presentation subscale included 7 items rated on a five-point Likert scale from strongly disagree (1) to strongly agree (5). Some example items included “I understood the exercises in the parenting toolkit” and “I trust the information in the parenting toolkit”. In this subscale, some items were adapted to refer to specific therapeutic elements of the intervention (e.g., “I understood the metaphors in the parenting toolkit (e.g., tree metaphor, passengers on a bus, beach ball etc.). These changes were informed by the findings of PAR workshops. The importance of breaking down complicated and abstract concepts was raised in the intervention development feedback workshops (see section 8.1.3.). One item from this subscale was removed (“I find the images on the website distressing”) based on feedback from public involvement representatives that this was irrelevant due to the nature of the images used in the design of the materials. This subscale also mapped onto the intervention coherence component of TFA.

User interaction with materials

The survey also contained questions to gather data on how participants interacted with the intervention materials. Participants were asked to provide an approximation of the total time they had spent viewing the materials and which sections of the materials they engaged in and which they had found the most helpful. They were also asked to indicate how often they might use the intervention materials if they were freely available.

To assess perceived burden (see TFA Figure 24), participants were asked to rate the length of the intervention materials using the following multiple-choice item: “Did you feel the length of the intervention was 1) too long, 2) too short, 3) just right”.

NHS Friends and Family Test

Items from the NHS Friends and Family Test (FFT; NHS, 2013) were included to gain an understanding of parents' overall experience of the intervention and whether they would recommend it to others. These items were "Overall, how was your experience of the parenting toolkit?" and "How likely are you to recommend the parenting toolkit to friends and family if they need support?". These items also mapped onto several concepts of the TFA: "affective attitude" and "perceived effectiveness".

Open-ended questions

Open-ended qualitative questions (e.g.: Is there anything about the parenting toolkit that you think could be improved?") were included throughout the survey to provide parents with a space to elaborate on their answers and share their experiences in their own words. These data were then used to enhance and clarify the quantitative data.

8.2.3. Research ethics

An ethics application was submitted to the Faculty Research Ethics Committee and returned with a few conditions to be addressed. For discussion of general ethical considerations of the PhD see section 3.8. Although ethical approval is typically not required when conducting Patient and Public Involvement (NIHR, 2021), an ethics application was submitted including both the PAR work and the acceptability study was submitted to the Faculty Research Ethics Committee and was returned with a number of conditions to be addressed. For discussion of general ethical considerations of the PhD see section 3.8.

This decision was taken due to the exposure of the public involvement representatives to the intervention materials. The intervention materials include therapeutic elements which required representatives to reflect on difficult thoughts, feelings, and experiences. Previous intervention development research with this population has reflected on the importance of treating informed consent as an ongoing process when reviewing therapeutic content (Zucchelli et al., 2021). Consequently, the intervention materials, the PAR workshops and the acceptability study protocol provided users with all the relevant information about the content of each section. This ensured that PAR representative and participants could provide fully informed consent throughout the review and feedback process, in line with ethical guidelines with psychological research (BPS, 2021).

8.2.4. Recruitment

The member organisations of the Appearance Collective were contacted via email and asked if they would advertise the present study to their members. The study was also advertised on the CAR social media pages and participant pool. To attempt to broaden the range of characteristics (e.g., gender, ethnicity, family structure), numerous strategies were employed. Online parent organisations who support underrepresented groups such as single parents or fathers were contacted (e.g., Single Parent Advice and Support Services, 'Dad's corner' forums). To attempt to include a wider range of family structures within the sample, foster and adoption support and advice services and forums for blended families were also contacted to ask if they would be happy to advertise the study to their members. Potential participants were provided a link to a Qualtrics form where they could register their interest.

8.2.5. Participants

Twenty-two participants took part in the acceptability survey. All (n=22) identified as female and their child's mother. The mean age of parents was 37.41 years (SD=7.33). Most identified as White British (n=13). Parents reported that the mean age of their child with a visible difference was 8.25 (SD=5.31). It was also reported that 14 of these children identified as female and 8 as male. The majority of children had a birthmark condition (n=9), with the next largest groups being craniofacial conditions (n=4) and limb differences (n=4). Three children had alopecia and there was one parent of a child with a burn injury and one parent of child with a skin condition. See Table 22 for a summary of demographic information.

Table 19: Demographic information of parent participants

	Mean	SD
Parent age	37.41	7.33
Child age	8.25	5.31
	N	%
Parent gender		
Female	22	100
Male	0	0
Child gender		
Female	14	63.6
Male	8	36.4
Relationship to child		
Mother	22	100
Father	0	0
Ethnicity		
Asian Chinese	1	4.5
White British	13	59.1
White European	3	13.6
White Irish	1	4.5
White Other	1	4.5
White Scottish	3	13.6
Child condition/injury		
Alopecia	3	13.6
Birthmark	9	40.9
Burn injury	1	4.5
Craniofacial condition	4	18.2
Limb difference	4	18.2
Skin condition	1	4.5

8.2.6. Procedure

Parents who expressed an interest in participating were contacted via email with a copy of the participant information sheet and consent form. Once participants returned these documents to the PhD student, they were provided with a copy of the intervention materials and a link to the acceptability survey. Parents were instructed to spend time reading and reviewing the materials and then complete the survey. Once participants clicked on the link to the survey, they were guided through a series of multiple choice and open-ended questions regarding their experience of using the parenting toolkit (see Appendix L). If a parent had not completed the feedback survey within one week of receiving the materials, they were sent a reminder email. At the end of the survey parents were thanked for their time and contribution and given the opportunity to enter a prize draw to win a £50 shopping voucher.

8.2.7. Data analysis

Quantitative data were analysed using SPSS Version 28. Descriptive statistics were calculated from each subscale of the eHIQ. Frequencies were calculated for the FFT and user interaction data. Subscale totals and means were calculated using an algorithm published in the eHIQ scoring guidelines (Kelly et al., 2015). The existing health literature has quoted a cut off for a positive score: ≥ 65 out of a maximum of 100 (Talboom-Kamp et al., 2020). The qualitative data were analysed using deductive content analysis (Elo & Kyngäs, 2008). Deductive content analysis has been previously described in detail in section 6.2.7. Content analysis is an appropriate approach for the present study because the qualitative data will be used to enhance the understanding of the quantitative data. Therefore, the concepts assessed in the quantitative data (e.g., information and presentation) can act as an existing conceptual framework for deductive analysis.

8.3. Results

A sample of 22 parents of children with a visible difference took part in an acceptability survey to provide feedback on the parenting intervention materials. The following section of this chapter will present a summary of the descriptive quantitative analysis and the deductive qualitative content analysis.

8.3.1. User interaction

Participants reported engaging with the intervention materials for a mean total duration of 54.32 minutes (SD = 51.14, Range = 15 – 240). A summary of participant engagement in the various sections of the intervention materials can be found in Table 20. Ten participants reported that they found the content related to “Talking about your child’s condition or injury" the most helpful section of the intervention. Eleven participants reported actively engaging in the activities and exercises. Most participants (n=10) stated that they would be most likely to use the intervention materials "as and when" they needed them or in response to a specific challenge, whereas others felt that they would use them more regularly: monthly (n=3) or weekly (n=4). Several parents (n=3) also shared that they wished that they had had access to the materials when their child was younger, as they would have used them more at this time. Most parents reported that the length of the materials was "Just right" (n=15), however several stated that the materials were "Too long" (n=7).

Table 20: Parent-report engagement with intervention materials N = no. of parents viewed each section.

Intervention section topic	Parent-reported engagement N (%)
Introduction (why support parents?)	18 (82)
What are difficult thoughts and feelings?	20 (91)
Helper skills	18 (82)
Being present	17 (77)
Knowing your values	19 (86)
Talking about your child's condition or injury	21 (95)
Other people's reactions	21 (95)
Staying informed	17 (77)
Finding the right support	19 (86)

8.3.2. eHealth Impact Questionnaire

After reviewing the intervention materials, parents responded to questions from two subscales of part 2 of the eHIQ questionnaire. Possible overall scores on each subscale

Table 21: A summary of the descriptive statistics for the eHIQ subscale scores, ≥ 65 cut off for a positive score.

Subscale	Mean	Standard deviation
Confidence and identification	81.94	12.27
Information and presentation	86.85	12.68

ranged from 0 to 100, with higher scores indicating more positive experiences (Kelly et al., 2015). A summary of the descriptive statistics for each subscale can be found in Table 21.

8.3.3. Friends and Family Test

All participants reported that overall, they either had a “good” or “very good” experience when engaging with the intervention materials. All participants also reported that they would either be “likely” or “very likely” to recommend the intervention to another parent of a child with a visible difference. See Table 22 for a summary of these findings.

Table 22: Summary of FFT results for 22 parent participants

Survey item	Frequency (%)
Overall, how was your experience of the parenting toolkit?	
Good	7 (32)
Very good	14 (64)
Did not report	1 (4)
How likely are you to recommend the parenting toolkit to friends or family if they need support?	
Likely	4 (18)
Very likely	17 (78)
Did not report	1 (4)

8.3.4. Content analysis

Two categories were identified in the qualitative data: 1) Confidence and identification, and 2) Coherence, presentation, and layout. These categories were informed by the subscales of quantitative measures used in the present study. Each category and the subsequent subcategories will be discussed in greater depth in the following section. A “following a thread” approach was used to integrate the findings from the quantitative and qualitative data sets. A description of this method can be found in 6.3.4. When discussing the findings, guidelines around quantifying language were adhered to (Hill et al., 2005). “All” refers to all participants, “most” refers to more than half, “some” refers to less than half but more than two. A visual representation of the categorisation matrix and exemplar quotes for each category can be found in Appendix L.

Confidence and identification

All parents within this sample (n=22) discussed the extent to which they found the content of the intervention material to be relevant and identified with the topics. Some of these (n=10) provided positive feedback on the topics included and felt that the intervention provided comprehensive support to parents. Some (n=9) reported that the content closely reflected their own experiences of caring for a child with a visible difference and the information and exercises were relevant to their experiences, past and present. Within these parents, six also felt the materials would be most beneficial early in a parent’s journey (e.g., after a child was born or immediately following an acquired appearance difference). Two parents also shared their continued need for support later in their child’s life. In addition, one parent suggested that more direct quotes from parents would help users identify with the content. Two parents also explicitly expressed that they had enjoyed using the materials.

Coherence, presentation, and layout

Most parents (n=18) also discussed the coherence, presentation, and layout of the intervention materials. Thirteen reported they understood the content (e.g., language, sentence structure). Most (n=11) identified no problems with comprehension and stated that the content was clear and well explained. However, two parents described difficulties fully understanding some of the metaphors used to explain ACT processes (e.g., beach ball metaphor, passengers on a bus). Two parents discussed the use of colour and illustrations within the materials. They stated that the use of illustrations and colour was eye-catching and helped to balance the amount of text on the page, whereas two others felt the colour blocking within the design was confusing.

Some parents (n=9) commented on the length of the materials. Five stated that some sections were too long and could be condensed. However, others (n=4) felt that the length of the materials was appropriate and that they benefited from the amount of information included. Some (n=5) discussed the format of the intervention delivery. Four parents felt that they would like the option of having the materials in a physical printed format, rather than being digital only. One parent also suggested that due to the therapeutic nature of the content, it might be helpful to have more human interaction, such as an audio recording of someone reading the materials.

8.4. Discussion

This study aimed to assess the feasibility and acceptability of a self-administered ACT intervention for parents of children with a visible difference. The study employed a mixed methods survey design to assess parents' experiences of engaging with the intervention materials. User-reported interaction data indicated a high level of engagement with the different sections of the intervention. Participants reported high mean scores on measures relating to identification with the content and confidence using the materials. Parents' scores on information and presentation also highlighted that they found the content to be clear and well presented. The qualitative data provided greater detail of participants' experiences. This included clarification on any content that they struggled to understand and greater elaboration on opinions about length of the intervention materials. These findings will now be discussed in relation to acceptability literature and the previous findings of this PhD.

8.4.1. User interaction

Parents reported high levels of engagement with the intervention materials. Between 77 and 95% of the sample engaged with each section. Parents were not instructed to read the entire intervention or engage with each section, suggesting that this engagement was guided by their own interest and free choice. Parents were asked to report which informational sections and exercises they thought were most helpful. Many parents identified the sections on "Talking about your child's condition or injury" to be most helpful. This aligns with the current literature in the field and the previous findings of the PhD. Recent qualitative studies have found that raising appearance-focussed discussions is a primary concern of parents of children with a visible difference (Feragen et al., 2021; Zelihić et al., 2021). The mixed methods findings of the present PhD also indicate that parents lack confidence about how to have these conversations. The second study of the PhD also indicated that good parent-child communication about their child's condition could be a protective factor against parent psychological distress. The user-reported activity further supports the evidence that communication about visible difference is a significant concern for parents. To the PhD student's knowledge, this is the first evidence-informed parent resource which directly focuses on talking to their child about their visible difference. Consequently, the present intervention materials address a previously unmet support need for parents of children with a visible difference.

8.4.2. Friends and Family test

Parents also completed the NHS FFT to provide feedback on their overall experience of the intervention materials. Sixty-four percent of parents rated their overall experience as "very good" and 32% of parents rated their experience as "good". In addition, 78% of parents reported that they would be "very likely" to recommend the parenting toolkit to other parents, whilst 18% reported that they would be "likely" to do so. These findings closely reflect those of previous acceptability testing conducted on an evidence-informed website for parents of children with burn injuries (Heath et al., 2019). These findings suggest that the intervention materials were acceptable to parents in the present study.

8.4.3. Identification with content

Parents reported a mean score of 81.94 out of a maximum 100 on the confidence and identification subscale of the eHIQ. This suggests that parents identified strongly with the content and felt confident in their ability to support their child after viewing the materials. The materials reflected and addressed their experience and the challenges. They also stated that the content increased their confidence in their ability to manage these challenges. These findings support the theme identified the PAR workshops, “Meeting support needs”. In this theme, parents and professionals indicated that the content included in the intervention reflected their experiences and observations of parent support needs. Therefore, both groups suggest that the content of the intervention is acceptable to parent users.

These findings align with several of the components of TFA (Sekhon et al., 2017). Firstly, parents expressed in both the quantitative and qualitative data that they felt the intervention materials would be helpful to themselves and other families. Based on their experience of viewing the materials, they believed they could be effective. This aligns with the TFA construct of “perceived effectiveness” (Sekhon et al., 2017). Secondly, parents reported feeling confident in their ability to support their child. This aligns with the TFA construct of “self-efficacy” (Sekhon et al., 2017). The second study of this PhD also found self-efficacy specific to caring for their child with a visible difference to be significantly positively associated with positive affect. This suggests that the content of the support materials meets the needs of parents in this sample and is perceived to be acceptable.

Additionally, this subscale also asked parents to reflect on whether the content of the intervention aligned with what is important to them and their family, i.e., their values. This aligns with the TFA construct of “ethicality”, defined as “the extent to which the intervention has good fit with an individual’s value system” (Sekhon et al., 2017, p.8). The concept of values and value-based action is also at the core of ACT (Hayes et al., 2006). This also links to a theme from the PAR phase: “Getting parents on board”. This theme described the importance of building trust and engagement with parents by demonstrating that the authors of the intervention understand their experiences. These findings suggest that the parents in the present sample closely identified with the intervention content and found the materials to be acceptable.

The qualitative data suggest that several parents in the present sample (n=6) would have welcomed the intervention materials at the beginning of their journey with their child’s visible difference. This supports condition-specific literature with both mothers and fathers (e.g., Heath et al., 2018; Stock & Rumsey, 2015) and the cross-condition findings of the PhD which indicate that parents are particularly in need of psychosocial support in the early days of their child’s condition or injury. Further work to explore effectiveness of the intervention in parents of young children or children who have recently acquired a visible difference would be beneficial.

8.4.4. Information and presentation

Parents reported a mean score of 86.85 out of a maximum 100 on the information and presentation subscale of the eHIQ. Overall, parents felt the presentation of the content of the intervention was suitable. This subscale included items about how accessible and easy the content was to understand. These findings align with the TFA component “intervention coherence” defined as “the extent to which the participant understands the intervention, and the way the intervention works” (Sekhon et al., 2017, p.9). Intervention coherence also links to several themes from PAR phase of intervention development “Accessibility of format” and “Communicating complex concepts”. Both themes discussed the importance of ensuring that the complex concepts within the intervention materials are clearly communicated using accessible language and other techniques such as real-world examples and metaphors. The quantitative and qualitative data in the present study indicated that most parents felt that the content of the intervention was clear and easy to understand. Only two parents reported challenges understanding some of the metaphors used to illustrate ACT concepts. These findings suggest that the content of the intervention was clearly presented and easy to comprehend.

Parents also provided feedback on the length of the intervention materials. Seven of the 22 participants reported that the materials were “too long”, whilst 15 reported that the length was “just right”. This split in feedback was also observed in the qualitative data, with five participants reporting concerns about the length, whilst four others stated that they felt the materials were the right length. Of the parents who felt that the materials were the right length, several also acknowledged that there was a lot of information included. However, they felt the information was important to include and that they benefitted from engaging with it. This feedback aligns with the “burden” and “opportunity costs” within the TFA. Both components consider the possible costs (e.g., time, energy, effort) of engaging in an intervention.

Furthermore, these findings were also reflected by a sub-theme from PAR phase: “There’s no getting away from the fact that it’s long” (See Appendix K). Similar to the present findings, parents and professionals in the PAR workshops reflected on the length of the content and highlighted that there was a lot of important topics covered. In addition, parents in the PAR phase suggested methods to help users find the information they wanted. In the present study, the majority of parents reported that they would use the materials in response to particular challenges and when needed. To balance potential costs to participants with benefits of the resource, further changes could be made to allow parents to easily find specific topics and dip in and out of the intervention materials. This could reduce burden (e.g., time and effort) of engagement, whilst continuing to provide parents with valuable information.

8.4.5. Limitations

The present sample may not be typical of those who may require psychosocial support, as parents were self-selecting and volunteered to review the intervention materials. For example, the mean age of children was 8.25 years (SD: 5.31 years). The qualitative data indicated that some of these parents had been living with their child’s condition for several years and suggested that this kind of support may be more appropriate when the child is younger or immediately after the visible difference is acquired. As a result, some feedback may not represent the experience of all parents who may choose to use the materials were they freely available.

Due to the self-administered nature of the present intervention, parents varied in their interactions with the intervention materials. The data indicated a large range of viewing times (15-240 minutes), which suggests parents may have had varying levels of exposure to the intervention sections. Existing literature exploring psychosocial intervention implementation has discussed a phenomenon called a “dose response”, wherein greater exposure to the intervention content is associated with improvement in outcomes. For example, the dose response has been found in a RCT exploring the effectiveness of an ACT-based mobile intervention (Mattila et al., 2016), where increased number of completed sessions was associated with greater improvements in psychological flexibility. Consequently, any future work to test the effectiveness of the intervention materials should include clear guidance for participants about which key areas of content to access. More in-depth data should be collected about participant engagement to monitor for differences relating to a dose response.

8.5. Conclusion

The present study aimed to assess the feasibility and acceptability of a self-administered ACT-based intervention for parents of children with a visible difference, developed in response to the mixed methods findings. Parents reported that its content resonated with their lived experiences of caring for a child with a visible difference and materials were in line with their values. High scores related to presentation of the information also indicated that parents perceived the information to be easy to understand and clearly presented. Parents felt the intervention materials would be most beneficial for parents whose child has either just been born with a visible difference or recently after their child has acquired a visible difference. Additionally, the qualitative data suggested that some parents had concerns about the length of the intervention. However, parents felt that further refinement of the design of the materials would reduce potential burden or costs (e.g., time and energy) of engaging with the intervention, and increase benefits. This work has not yet been completed as it was out of the scope of the timescale of this project. The following and final chapter of the PhD thesis will explore and reflect on the overall conclusions from the research and intervention development process and discuss possible directions for future work in this area.

Chapter nine: General discussion and final reflections

This final chapter discusses the research conducted, the application of the findings, and the intervention output produced, in the context of the existing literature and theory. The strengths and weaknesses of the research methods are reflected upon and future directions for the field are considered.

9.1. Summary of research process

This PhD set out with a broad scope to explore the experiences and support needs of parents and carers of children and young people with a visible difference. Upon review of the literature, it became clear that research with parents of children with a visible difference was scarce and limited in its approach. At the beginning of the PhD (2018), much of the parenting literature in this field was condition-specific and confined to only a few appearance-affecting conditions and injuries (e.g., CLP and burn injuries; Heath et al., 2018a; Nelson et al., 2012; Stock & Rumsey, 2015). The experiences of many parents of children with a visible difference, particularly those with rarer conditions, were under researched and their support needs unknown. Furthermore, there was very little evidence-based support for parents of children with a visible difference. More recently, the range of conditions and injuries included in parenting research has begun to broaden (e.g., limb differences, Oliver et al., 2020; alopecia, Putterman et al., 2019) and this, alongside the investigations conducted within this program of work, has progressed the understanding of the experiences of parents.

Having worked in an NHS service with parents of children with rare appearance-affecting conditions, I was aware that many of the challenges discussed in the existing condition-specific research were also relevant to the issues discussed in clinic appointments. A review of the literature suggested that children and young people with a wide range of visible differences experience similar psychosocial challenges (Jenkinson et al., 2015).

Therefore, a cross-condition approach was taken to investigate whether parents of children with a visible difference had common experiences and challenges, regardless of the cause or nature of their child's condition or injury. My positionality as a previous employee of charitable organisations and the NHS gave me clear insight into health service resources and the kind of support that might be implemented effectively in these settings. Therefore, my research took a pragmatic approach to developing a knowledge base of the experience of this population and to better understanding of the gaps in support. It was important to me that my research work had practical applications, improved evidence-informed support provision, and added to the work of existing charitable organisations (e.g., Changing faces: https://www.changingfaces.org.uk/).

The parenting intervention developed based on the mixed methods findings of this PhD is novel in its evidence-informed cross-condition approach. The intervention materials also included the integration of an innovative self-administered ACT-based therapeutic approach with practical advice and guidance. This intervention could make a significant contribution to the support available to parents of children with a visible difference. To reflect fully on the development of this output and the generation of new knowledge in the field, it is important to first review the PhD research process. The MRC guidelines for development of complex interventions (Figure 21) will be used to guide this reflection.

Firstly, studies 1 and 2 were associated with the "develop intervention" phase of the MRC guidelines. These studies collected data using a mixed methods design, utilising qualitative interviews and focus groups (study 1) and an online survey (study 2). The findings from these studies were then integrated using a triangulation protocol (Farmer et al., 2006). These findings formed the evidence base for understanding cross-condition experiences and support needs of parents. Based on these novel findings, intervention materials were drafted and written with support from experienced researchers and clinicians within the field. Using a collaborative Participatory Action Research approach, these draft materials were discussed and reviewed by parent representatives and health and support clinical advisors. The PAR approach utilised group workshops to collate feedback on materials and inform the continued development of the intervention. Parents and health and support professionals provided suggestions for changes. Following iterative changes made in line with findings from the PAR workshops, study 3 moved into the feasibility stage of intervention development and aimed to assess the acceptability of the content and format on the materials. Acceptability data was collected using an online survey design.

Overall, the intervention development combined a theory and evidence-based design and a partnership approach (See section 3.12. for discussion of taxonomy of intervention development; O'Cathain et al., 2019). Initial decisions regarding the content and the therapeutic approach of the intervention were based on the mixed methods findings of the first two studies of the PhD. However, decisions regarding topics covered in the practical advice section, language, and presentation were informed and guided by collaborative discussion with parent user representatives and professional advisors.

9.2. Reflections on research methods

This section of the chapter reflects on specific aspects of the methods used throughout the PhD and considers the strengths and weaknesses of the design and conduct of the programme of work.

9.2.1. Mixed methods design and integration

A pragmatic mixed methods approach was employed for the studies in this PhD program of work. This enabled the research design to prioritise the utility of research and focus on practical problem solving (Rorty, 1999), using the most appropriate method to serve the aims of the research (Johnson & Onwuegbuzie, 2004). Overall, the cross-condition mixed methods findings are novel in the field of visible difference research. The use of mixed methods enabled the creation of new knowledge and allowed development of further understanding of the experiences and support needs of parents. Furthermore, the pragmatic approach to this research area allowed for the creation of an innovative intervention and has contributed to the evidence-informed support provision for parents.

The data from studies 1 and 2 were integrated using a rigorous and systematic approach (see Chapter 7 for description of integration process). An ongoing debate regarding the implementation and transparency of mixed methods research is the process of how to combine data and paradigms to draw overall conclusions (Johnson & Onwuegbuzie, 2004). The present PhD considered integration methods throughout the design and conduct of the research. Several methods of integration were utilised at various stages of the research process. This included the use of the triangulation protocol (section 7.3.2.; Farmer et al., 2006) within study 1 and to integrate the two overall data sets of studies 1 and 2 and "following a thread" (see section 6.2.7.6.; Moran-Ellis et al., 2006) within studies 2 and 3b. Interpretative rigour and inference transparency are both major elements of the quality framework for mixed methods research (O'Cathain, 2010). Therefore, the rigour demonstrated within the integration processes is a major strength of this mixed methods PhD.

Despite the strengths of this mixed methods approach, it is important to consider any challenges. Firstly, mixed methods require researchers to possess the knowledge and expertise in both qualitative and quantitative methods and learn to move between the two effectively (Johnson & Onwuegbuzie, 2004). As previously discussed, (see section 3.6.) methodological purists posit that researchers should always work within either a qualitative or quantitative paradigm (Johnson & Onwuegbuzie, 2004). Although, moving between paradigms throughout the PhD was challenging, the process of conducting mixed methods demonstrated the utility of a pragmatic approach in answering "real world" questions without the constraints of being tied to strict principles of traditional research paradigms (Feilzer, 2010). Another challenge of working with mixed methods can be that findings from different data sets may conflict or contrast. Case studies of mixed methods research with conflicting findings have stated that although this is a challenging position, disagreement within data may be a consequence of one paradigm tapping into a perspective or facet of experience that could not be observed through the other (Reams & Twale, 2008). Rather than being a problem for researchers to solve, conflicts in mixed methods data lead to deeper understanding of the multifaceted and nuanced phenomena which psychological research aims to understand.

9.2.1.1. Sample characteristics

The diversity of demographic characteristics was reflected upon and considered throughout this programme of work. This section discusses the steps taken to broaden the samples of the present PhD, limitations within samples, and the possible implications of these.

Gender and parenting role

The majority of samples in parenting research in the visible difference field have an overrepresentation of mothers (e.g., Heath et al., 2018; Hlongwa & Rispel, 2018; Koot et al., 2000; Razera et al., 2017). This is also true of parenting research (e.g., Macdonald et al., 2010) and psychological and health research more broadly (e.g., Ryan et al., 2019). Although fathers were present in the samples of study 1 (n=11 out of total n=45) and study 2 (n=23 out of total n=209), there remained a significant gender imbalance within these samples. However, despite efforts to recruit fathers (e.g., contact specific support groups and forums for fathers), there were none present in the samples of study 3.

After noting a lack of representation of fathers’ perspectives in the existing literature, the recruitment plans for each study of the PhD included strategies to reach more fathers. These strategies included: snowball sampling through female partners, contacting support and advice groups specific to fathers, and attending charity events where both parents were present. Snowball sampling through female social contacts has previously been effective for increasing male recruitment (Ryan et al., 2019). However, this became more challenging and a less effective strategy after face-to-face events were cancelled due to the

COVID-19 pandemic. This was reflected in the present PhD, as both studies 1 and 2 utilised snowball sampling. Study 3 also recruited a female and male parent couple through snowball sampling, however this couple later dropped out of the study due to family commitments. Research from the visible difference field (Stock et al., 2020) and the broader literature (Jia et al., 2016) has illustrated that men and women have different coping strategies in response to parenting challenges. The broader health literature has also identified that fathers have different caregiving roles to mothers. For example, mothers are often the gatekeepers of various forms of care (e.g., medication and healthcare appointments; McGrath & Chesler, 2004). This has important implications for intervention development. Although the data collected in study 3 provides important insights into the acceptability of the intervention, this may not be generalisable to fathers. Further acceptability testing and piloting with male caregivers may be required before the intervention materials can be evaluated.

Sociocultural background

The PhD student employed strategies to attempt to broaden the sociocultural diversity of the participants of the PhD. This included contacting organisations across a wide range of geographical areas around the UK and including family support organisations based in lower SES regions. However, despite these efforts there were limitations within these samples. Literature on recruitment and retention of low-SES parents recommends the development of community partnerships. Although more challenging during the COVID-19 pandemic, this may have improved recruitment rates of families from a wider range of backgrounds (Baucom et al., 2018).

The majority of participants identified as White British and highly educated. Although not unusual in psychological research (Muthukrishna et al., 2020), this does have important implications for the findings and outputs. The lack of cultural diversity within the sample is important to reflect upon because parenting theory has indicated that individuals from different cultural backgrounds demonstrate variations in parenting behaviours (Lamm & Keller, 2007). Families from black and minority ethnic backgrounds may also be subject to further social inequality through the experience of intersectional stigma (Turan et al., 2019; see section 9.4.), which may negatively impact on psychosocial adjustment. As such, findings from a sample of predominantly White British parents may not generalise to the experiences and support needs of parents from other cultures.

Additionally, the majority of the participants were also educated to first degree level or above, possibly indicative of higher socioeconomic status (SES). As a result, the representation of different SES groups was limited. Data regarding employment status, housing, income, or other variables indicative of social class were not collected, which further limits the conclusions that can be drawn across SES groups. As previously discussed, (see section 4.5.) parents from higher SES backgrounds may have access to additional coping resources (e.g., financial support) which could aid in coping with challenges related to their child's condition or injury. For example, many conditions or injuries require regular hospital appointments which can be costly in both time and money (e.g., Hlongwa & Rispel, 2018). Parents from a higher SES background may have more resources to cope with associated practical challenges, such as the cost of travel and greater flexibility to take time off work. A possible reason for this imbalance in SES within the sample could be the reliance on online recruitment due to COVID-19 (see Appendix E).

Digital exclusion can be associated with lower levels of education and higher rates of unemployment (Helsper & Reisdorf, 2017). Therefore, online recruitment may have inadvertently excluded parents from socioeconomically disadvantaged families. This is also important to reflect on in the context of intervention development, when considering the mode of delivery (e.g., online vs hard copy materials). Creating offline, as well as online, formats could increase accessibility of the intervention materials for groups who may not have consistent access to internet enabled devices. High levels of attrition are common in parenting interventions, particularly within families from a socially disadvantaged background (Katz et al., 2001). Consequently, it is important to ensure that interventions are acceptable to parents from a range of SES prior to implementation.

Family structure

After becoming aware of a bias towards “traditional” two parent families in the sample of the first study of the PhD, strategies were implemented to broaden the diversity of family structures included within the research. These included contacting support organisations and forums which specifically support single parents, blended families, and adoptive and foster families. Consequently, there was some representation of single parents and blended families within studies 2 and 3. The broader paediatric illness literature has identified that the experience of single parents of children with chronic illness is understudied (Brown et al., 2008). Although non-parent carers have been included in existing work on parents and carers of children with visible differences (e.g., Heath et al., 2018), traditional mother-father relationships still make up the majority of samples. Where non-parent carers have been the main focus of research, it has been found that these individuals also have unmet support needs. A qualitative study of 12 grandparents of children born with CLP found that participants experienced difficult emotions, particularly during the time of diagnosis, and had ongoing concerns about the child’s treatment and future social challenges (Guest et al., 2019). Therefore, the inclusion of a range of family structures within this research provides an important contribution to the visible difference literature. More work could be done to further diversify samples in future research, such as greater inclusion of non-parent carers.

9.2.2. Inclusion of public involvement

Patient and Public Involvement (PPI) was a central element to the research conducted throughout this PhD. See section 3.11. for a full definition and description of PI in the present PhD. PPI was present in each study and parent user representatives and health and support professional advisors were involved in the development of the intervention materials, using a Participatory Action Research model (see section 8.1.3.; Baum et al., 2006). This collaborative approach to intervention development facilitated shared power and decision-making between the PhD student, the user representatives, and advisors. These experts by experience were able to guide intervention development discussions and ensure that the content and design of the intervention was meaningful and sensitive to the needs of families (Shen et al., 2017).

There can be challenges that arise from public involvement. Some key challenges identified by a scoping review of current practices included: time required to build relationships, monetary costs, and lack of clarity around public involvement roles (Shen et al., 2017). In the present PhD, the relationships with the core members of the parent advisory group were built six months prior to starting data collection for the first study. By the intervention development stage of the PhD, these individuals had been engaged in the project for three years, trusted the PhD student and were familiar with the project. Although fostering these relationships was time-consuming, this enabled consistent and valuable parent involvement throughout the PhD. Monetary compensation was provided to all public involvement representatives. However, this was challenging on a limited PhD budget and emphasised the importance of budgeting public involvement costs from as early as possible in the research design process. To provide parents with clarity about roles in involvement work, the PhD student provided training on public involvement at the beginning of the PhD and then at key points throughout (e.g., recruitment of new public involvement represents and at beginning of intervention development). Overall, PI was an integral element of the PhD process, which provided valuable input to all studies and the intervention output.

9.3. Discussion of findings and relevant theory

As discussed by Belsky's (1984) determinants of parenting model (see section 2.4.1.), parenting is multi-dimensional and dependent on the parent, child, and social contexts. According to this model, a wide variety of complex and interconnecting factors can affect parent adjustment (Taraban & Shaw 2018). This theoretical position has been reflected in the design and findings of the present PhD. Notably, Belsky (1984) emphasises the importance of the parent-child relationship in coping. This is also considered in many other developmental theories and is a theme which has continued to run throughout the findings of this PhD and is at the core of the intervention content. Theory related to individual parenting styles (Baumrind, 1971), family systems theory (Broderick & Smith, 1979b), the bioecological model of human development (Bronfenbrenner, 2005), and Social Learning Theory (SLT; Bandura, 1986) all consider this process to be central to understanding parenting, and therefore understanding the development of the child. Insights from the mixed method findings also lend support to these theories of parenting. For example, these findings highlighted parent preoccupation with modelling adaptive coping responses and behaviours to their child. This echoes the interconnected nature of families that is posited by family systems theory (Broderick & Smith, 1979) and the bioecological model (Bronfenbrenner, 1971), and the impact these interpersonal and social environments can have on child development. Furthermore, these findings also suggest that parents may directly impact child behaviour through responses to parenting styles (Baumrind, 1971) and/or observation and imitation of parent behaviours (SLT; Bandura, 1986). The mixed method findings and relevant theory illustrate the important implications and applications of further understanding parents' experiences and support needs, to promote parent and child adjustment.

The importance of the parent-child relationship is also addressed in the opening section of the intervention materials. A metaphor which describes parent-child relationship as a growing tree, where the parent forms roots which then go onto to nourish the child's development (see Appendix K.), is utilised to communicate to parents that by seeking support from themselves, they can also obtain greater skills and resources to support their child. As highlighted in the findings of the PAR workshops (see Appendix K.), drawing on parenting theory and tapping into this parent-child link, may help to contextualise the impact of the parent-child relationship and the importance of parents taking care of their own support needs, so that they can then provide better support to their child. Parents in this study felt that the tree was a good analogy and one that resonated. Consequently, framing the intervention content around the parent-child relationship was considered acceptable to parent user representatives.

Studies 1 and 2 also discussed more distal influences of parent adjustment. The bioecological model of human development would describe these as systems within the exosystem or macrosytem (Bronfenbrenner, 2005). For example, healthcare systems may interact with the parent microsystem to pass on information about the child's condition or treatment. Studies 1 and 2 identified that knowledge and satisfaction with their child's care was associated with reduced negative affect and stress. Thus, this exosystem and microsystem interact and impact parent adjustment. The bioecological model does not explicitly account for interactions directly between these two systems. This model may not fully represent the adjustment in families for whom chronic health conditions are part of daily life and, as a result, health and social care contexts become more proximal to the parent microsystem. The model does however demonstrate interactions between different microsystems, known as mesosystems. One example of this is the interactions between parents and school. Study 2 found that increased parent-reported teasing was found to be associated with negative affect and stress. Therefore, these findings support the existence of interactions between microsystems which may impact parent adjustment.

As well as acknowledging the importance of the parent-child relationship, the intervention materials also recognise the importance of context, thereby taking a similar approach to the bioecological model (Bronfenbrenner, 2005). One example of this is the value-based action exercise which asks parents to consider how they might behave in line with their values in different contexts. Study 3 found that 86% of parents engaged with this content. Additionally, both parents and professionals gave positive feedback about the exercise. Therefore, this approach of contextualizing value-based action was considered acceptable by parents and professionals within these samples. This supports the context element of the PPCT (Person-Process-Context-Time) theory that underpins the bioecological model (Bronfenbrenner, 2005), and suggests that this model has utility for understanding parenting experiences in this population.

The intervention materials were designed to be a self-administered, low-intensity psychosocial intervention. As previously discussed (see section 8.1.2.3.), the CAR framework would categorise this type of support as a level 2 stand-alone intervention. The intervention is designed so that a parent can access and use the materials independently, without facilitation by a trained professional. However, professionals in the PAR workshops stated that they could see themselves using the intervention materials within therapeutic sessions within parents (i.e., as a Level 3 intervention). This indicates flexibility with respect to where the intervention may be positioned within the CAR framework.

Stepped care models have been adopted by the Department of Health (2001) to improve access to low-intensity forms of psychological therapy and improve mental health outcomes. The aim of the stepped care model in mental health treatment is to ensure the cost-effective use of limited healthcare service resources (Department of Health, 2001). Access to psychological therapy is restricted by the number of trained and available practitioners. Increased access to low level support and self-administered therapeutic tools (Kaltenthaler et al., 2002; Marks et al., 2003), allows specialised professionals to be available to deliver high intensity interventions, improving access and capacity for care overall. Similarly, the present intervention is design to provide foundational low intensity support. Given the limited psychosocial support available for this population (Morawska et al., 2015), this could make a valuable contribution to the stepped care provision for parents of children with a visible difference.

Stepped care models assume that low level self-administered interventions will be acceptable to individuals receiving support (Bower & Gilbody, 2005). Recent acceptability testing of an ACT-based mHealth intervention for individuals with a visible difference found that clinical stakeholders felt that self-administered therapeutic tools should not be a replacement for face-to-face resources (Zucchelli et al., 2021). However, parents in study 3 found the self-administered format of the intervention acceptable, although one parent suggested that more human interaction would be helpful (e.g., an audio recording of someone reading the materials). Overall, the research and intervention outputs support and align with parenting theory and a stepped care model of intervention delivery.

Although the PhD focussed solely on a visible difference population, these findings may have implications for the field of paediatric chronic illness more broadly. Alongside existing research (e.g., Hlongwa & Rispel, 2018; Jenkinson et al., 2015b; Magin et al., 2008), the mixed methods findings have indicated that children and young people with a visible difference, and their families, do experience appearance-related challenges. As discussed in section 2.2., these appearance-related challenges distinguish the experience of caring for a child with a visible difference from that of caring for a child with a less visible health condition. However, some of the challenges identified (e.g., attending hospital appointments, decision making about treatment, preparing a child to self-manage) could be applicable beyond the visible difference field. To some extent, parents of children with other health conditions may experience similar sources of anxiety. As a result, the ACT-based approach could be beneficial for parents of children with a wide variety of health conditions. ACT interventions have been found to be effective for parents of children with other health conditions including chronic pain (Wallace et al., 2016) and cerebral palsy (McMillan et al., 2020). Nevertheless, the transdiagnostic nature of the present PhD suggests that this could be an appropriate approach for families affected by a broad range of long-term health conditions.

9.4. Future directions

The present intervention development followed the process laid out in the MRC guidelines (Skivington et al., 2021). According to this framework, the intervention is in the feasibility stages of development. Acceptability of the content and the design has been assessed in study 3, which enabled the PhD researcher to recognise any further changes needed prior to moving onto the next stage of development: evaluation. The acceptability data indicated that some further changes to the design and format of the intervention materials should be considered. For example, a finding from study 3 was that some parents reported concerns about the amount of information included in the materials. It was suggested that although the content of the materials was important, the presentation of the information could be altered to facilitate easier navigation.

Parents and professionals both reported that having the choice of the materials in either electronic or hard copy would be beneficial. Online based support is a cost-effective solution to reach a large number of people in need of psychological support (Mook, 2014). This is particularly important given the limited face-to-face psychological services for individuals affected by visible differences (Williamson et al., 2018). Consequently, there is need for accessible self-help interventions which specifically focus on challenges related to visible differences (Bessell et al., 2010). In situations where access to healthcare is limited, it has been suggested that online access to support could help to address the treatment gap (Moock, 2014). Therefore, it is important that an online version of the parenting toolkit is available.

User representatives and professional advisors also proposed that having a choice of offline format would increase accessibility to this resource. This is supported by existing literature which explores the possible impact of digital inequalities and exclusion on health outcomes, when relying entirely on online resources (e.g., DiMaggio et al., 2004; Beaunoyer et al., 2020). For example, the current COVID-19 pandemic has highlighted how digital inequalities can lead to greater vulnerability to the virus, because individuals cannot access up-to-date online information regarding current protective measures (Beaunoyer et al., 2020). Older adults and socially and economically disadvantaged individuals are most likely to be at risk of the impact of digital inequalities (Yates et al., 2015). Therefore, producing an offline version of the parenting toolkit could increase reach and accessibility of this support resource. It is not within the scope of the PhD to integrate this next step of iterative changes; this should be carried out prior to the next stage of development.

There are several possible designs that could be utilised to evaluate the effectiveness of the intervention. Randomised Controlled Trials (RCT) have long been considered the "gold standard" of evaluation in evidence-based health research (Backmann, 2017). The random assignment of participants to different treatment conditions (e.g., intervention or control) and controlling for other variables provides a high level of confidence that it is the intervention itself that is responsible for differences between groups following the trial (Goodman et al., 2018). However, RCTs have received criticism for restrictive methodology and low external validity (Cartwright, 2007). RCTs are limited in their ability to provide context-sensitive conclusions regarding the effectiveness of complex health interventions (Marchal et al., 2012).

Alternative methods of evaluation include a realist approach, which is a pragmatic theory-driven enquiry which aims to answer the question "what works, how, in which conditions and for whom?" (Pawson & Tilley, 1997, p.48). The realist evaluation perspective sits within a critical realism paradigm, allowing for the exploration of causal mechanisms and the contexts in which they work (Kazi, 2003). This approach to evaluation has the advantage of increased transferability of findings (Kernick & Mannion, 2005). Literature has previously suggested that there is a lack of clarity regarding practical methods of conducting this kind of evaluation (Marchal et al., 2012; Porter, 2015; Wong et al., 2016). Other research outlines realist protocols and proposes that realist approaches can be a suitable for assessing healthcare initiatives (Coles et al., 2017). Overall, the current literature indicates that a realist approach to evaluation may provide an assessment of efficacy with greater transferability and external validity. Further consideration is required to identify the most appropriate methods and outcomes to evaluate the effectiveness of the present intervention.

As discussed previously, the samples of the research of the present PhD were limited in their diversity. Future research should aim to understand the experiences of more diverse groups of parents, including the intersectional experiences of individuals who identify as members of minority groups underrepresented in the existing literature. Stigmatised identities (e.g., appearance difference, low SES, minority ethnic groups) are often considered in isolation. However, many of these identities intersect and result in compounded stigma and discrimination (Turan et al., 2019). Existing evidence suggests that intersecting forms of stigma can influence mental and physical health and health behaviours in complex ways (Turan et al., 2019). There is currently limited research exploring intersectional stigma, including a lack of established appropriate instruments and research methods (Logie et al., 2021). Further work is required to develop effective methods to conduct intersectional research within the health research more broadly, as well as within the visible difference field.

Greater inclusion of fathers and other male caregivers would be beneficial to the field. One approach to including more male participants in health research could be to carefully consider recruitment techniques. Literature on male recruitment to health research suggests that the content and design of study advertisement and research materials can be unintentionally biased towards female engagement. An online mental health study found that adverts that utilised language associated with “strength” resulted in a higher recruitment rate when compared to adverts that focused on language related to “mental health” (Choi et al., 2017). A scoping review also found that tailoring language and communication style to conform to masculine norms (e.g., use of humour, informal language, alternative language for therapeutic work such as “upskilling”) can facilitate male engagement in psychological treatment (Seidler et al., 2018). Future work in this area should consider tailoring research advertisements, materials, and outputs (e.g., interventions) to promote greater engagement from male caregivers.

Additionally, the majority of research in this field is conducted with White samples from westernised countries. The limited research from non-western cultures has demonstrated variation in the experiences of parents of children with a visible difference (e.g., differences in causal beliefs, Olasoji et al., 2007). Consequently, it is important that future research includes parents and families from a wider range of ethnicities. This would provide insight into the experiences of parents from a variety of racialised groups and better understand whether existing support would meet the support needs of those living in different cultures. Public involvement representatives from underrepresented groups, such as male caregivers and parents from minority ethnic backgrounds, could provide insight into how best to design and implement research which appeals to and engages individuals from a broader range of backgrounds and experiences.

Furthermore, with greater understanding of minority groups, it may be possible to adapt the intervention to be accessible and appropriate for parents from different backgrounds. Interventions targeting body image and appearance concerns have been translated and adapted for different cultures, both for the general population (e.g., Confident Me, Craddock et al., 2021; Garbett et al., 2021), and for individuals with visible differences (e.g., YP Face It, Van Dalen, et al., 2021b). Once undergoing linguistic translation and cultural adaptation it was found that these interventions were acceptable within these populations. For example, “Confident Me” (a body image intervention for young people) has been adapted for use in India (Garbett et al., 2021). The cultural adaptations made to this intervention included addressing culture-specific appearance concerns (e.g., skin colour dissatisfaction), including appropriate popular culture references and examples, including reference to differences in family structure (e.g., extended family), and simplifying language to aid comprehension. Alongside input from members of the community, similar adaptations could be made to the present intervention to examine acceptability and effectiveness in other cultures and communities.

Existing research suggests that some aspects of the experience of parenting a child with a visible difference may also impact non-affected children within the family unit. Research with parents of burn injured children have found that parents may also limit the social and emotional development of siblings of affected children, to prevent further harm (Horridge et al., 2010; Phillips et al., 2007). Non-affected siblings can also experience reduced time and attention from caregivers who are responsible for providing care to their affected child. A qualitative study of 31 carers of children with eczema found that time-consuming daily treatment regimens meant that other children in the family developed maladaptive behaviours (e.g., scratching their skin) to gain attention from their parents (Santer et al., 2013). This suggests that siblings of children with visible differences may also experience psychosocial challenges. Further research could explore the experiences of siblings and their support needs, to build a comprehensive understanding of the impact on the whole family unit.

9.5. Final reflections and conclusions

The original aims of the PhD were: 1) to explore the cross-condition experiences and support needs of parents of children with appearance-affecting conditions and injuries, 2) to investigate possible risk and protective factors for parental psychological distress, and 3) to develop support materials to meet the identified cross-condition needs of parents of children with an appearance-affecting condition or injury. Building an understanding of the experiences and support needs of parents was important to promote psychological wellbeing. Based on the experiences and risk and protective factors identified in the mixed methods findings, the content for an ACT-based parenting intervention was drafted and reviewed by parent user representative and professional advisors. Finally, the acceptability of the content and design of an intervention was assessed using an online survey. The findings from this survey indicate that parents found both the content and the presentation of the intervention acceptable.

Consequently, the rigorously developed, evidence-informed intervention output has been found to be acceptable by a sample of parents of children with a range of visible differences. To the author’s knowledge this is the first therapeutic self-administered cross-condition intervention for parents of children with a visible difference of its kind that addresses an existing gap in support. As discussed, access to low intensity interventions provide accessible support to individuals with less complex and lower risk. This allows for the limited resources of specialist practitioners to be focussed on individuals who require more complex intervention, thereby increasing access to support for all. Following evaluation and dissemination, parents could be signposted to this intervention by charitable organisations and healthcare practitioners, thereby relieving some of the pressure on these individuals and services. This intervention has the potential to make a significant impact to the support resources available. The next essential step in ensuring availability of this intervention is to design and conduct a full evaluation to examine effectiveness.

This thesis has provided a critical overview of the existing literature detailing the experiences and support needs of parents of children with a visible difference. This knowledge has been explored in the context of the broader paediatric chronic illness literature and parenting theory. A pragmatic mixed methods research approach was used to produce new knowledge in the field of visible difference. This PhD has also produced an innovative intervention output, which is novel within the field. This program of work has also resulted in a peer-reviewed publication and academic conference presentations and the findings have been shared in non-academic settings such as charity events, a podcast episode, and blog posts. This has facilitated the dissemination of new knowledge and has raised awareness of the experiences and support needs of parents within the visible difference community. Creating this body of work has been incredibly rewarding and it is hoped that the outputs described will provide future insight and support for parents and caregivers.

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Appendices

Appendix A: Research outputs from this PhD

Academic publications

Thornton, M., Harcourt, D., Deave, T., Kiff, J., & Williamson, H. (2021). “Have We Done Enough?” A Cross-condition Exploration of the Experiences of Parents Caring for A Child with an Appearance-affecting Condition or Injury. *Developmental neurorehabilitation*, 1-11.

This publication can be found at the following link:

https://www.tandfonline.com/doi/pdf/10.1080/17518423.2021.1901150

Conference presentations

Thornton, M., Harcourt, D., Deave, T., Kiff, J. & Williamson, H. (2021, July) *Promoting psychosocial adjustment in parents/carers of children with appearance-affecting conditions and injuries: an investigation of the risk and protective factors.* PsyPAG Annual Conference. Online.

Thornton, M., Harcourt, D., Deave, T., Kiff, J. & Williamson, H. (2021, July) *"Have we done enough? A mixed methods exploration of cross-condition experiences of parents of children and young people with appearance-affecting conditions and injuries.* Appearance Matters 9. Bristol, England.

Thornton, M., Harcourt, D., Deave, T., Kiff, J. & Williamson H. (2021, July) *Creating a safe space: the use of activity-orientated focus groups with visual aids to explore the experiences of parents of children with Congenital Melanocytic Neavus*. Division of Health Psychology Conference. Online.

Thornton, M., Harcourt, D., Deave, T., Kiff, J. & Williamson H. (2020, January) "*They become so much more": Activity led focus groups exploring the journey of parent of children with Congenital Melanocytic Neavus.* Qualitative Research Symposium, Bath, England.

Thornton, M., Harcourt, D., Deave, T., Kiff, J. & Williamson H. (2019, September) *Exploring the cross-condition experiences of parents caring for a child with an appearance altering condition or injury.* European Health Psychology Society Conference. Dubrovnik, Croatia.

Presentations at charity events

Thornton, M. (2021, July) *Experiences of parents and carers of children with appearance-affecting conditions and injuries.* Ectodermal Dysplasia Society Medical Advisory Board. Online.

Thornton, M. (2019, July) *Experiences of caring for a child with Congenital Melanocytic Naevus.* Caring Matters Now family conference (workshop). Wiltshire, England.

Thornton, M. (2019, June) *Research with parents at the Centre for Appearance Research.* Microtia UK Family Fun Day. London, England.

Charity publications

Thornton, M. (2020, September 9) Developing evidence-based support for parents of children and young people with a range of appearance-affecting conditions [Blog post]. Retrieved from https://www.alopecia.org.uk/blog/developing-evidence-based-support-for-parents-of-children-and-young-people

Thornton, M. (2020, August 24) Developing support for parents and carers of children with appearance-affecting injuries [Blog post]. Retrieved from https://www.cbtrust.org.uk/2020/08/24/developing-support-for-parents-and-carers-of-children-with-appearance-affecting-injuries/

Podcast episode

Thornton, M., Rycroft, T., Parnell, J., Craddock, N., (2021, June 10) Parenting a child with a visible difference (No. 60) [Audio podcast episode]. In Appearance Matters: the podcast! Soundcloud. https://soundcloud.com/appearance-matters/parenting-a-child-with-a-visible-difference

Appendix B: Descriptions of appearance-affecting conditions and injuries

Alopecia: Alopecia is an autoimmune condition which causes hair loss There are several types of alopecia: 1) alopecia areata, 2) alopecia totalis, 3) alopecia universalis, and 4) androgenetic alopecia (Lee et al., 2020). Alopecia areata is characterized by patchy hair loss on the scalp or other places on the body (e.g., facial hair). Alopecia areata can be transient and affects approximately 2% of the general population at some point in their lifetime (Lee et al., 2020; Pratt et al., 2017). Alopecia totalis results hair loss to the entire scalp and a recent meta-analysis has found that the pooled prevalence of this type of alopecia is 0.08% (Lee et al., 2020). Alopecia universalis is a more advance presentation than alopecia totalis and results in hair loss across the entire scalp, face, and body (Lee et al., 2020).

Burn injury: Burn injuries are the fifth most common cause of a non-fatal injury in children (World Health Organisation, 2016). Burn injuries are described as first-, second-, or third-degree based on the depth of the burn and how severely they penetrate the skin (Baker, 2012). First-degree burns only affect the outer layer of the skin and are characterized by redness and painful dry skin, but no blistering. Second-degree burns affect the epidermis and dermis layers of the skin and cause blistering and swelling which can be painful. Third-degree burns severely damage the epidermis and dermis, and can cause damage to underlying bones, muscles, and tendons.

Cleft lip and/or palate: Cleft lip and/or palate (CLP) is a congenital condition which affects approximately 1-2 people per 1,000 newborns (Sivertsen et al., 2008). CLP is caused by a disruption to the cell fusion process during early fetal development. (K. B. Feragen, 2012). The severity of the cleft depends on the timing of any issue that arise. A cleft may involve the lip and/or the palate and can be unilateral (affect either the right or left side) or bilateral. The primary treatment for this condition is surgery to repair the lip and palate, typically carried out in the first year of life. However, treatment can continue throughout childhood and into adulthood (Stock et al., 2015).

Congenital melanocytic naevus (CMN):CMN is a rare type of congenital pigmented birthmark which can vary in size and colour (NORD, 2019). CMN can have hair growth and the texture of the birthmark tends to be different to the rest of the skin (NORD, 2019). A CMN can be single of accompanied by multiple dispersed satellite neavi (NORD, 2019). Small single CMN are present in 1% of newborns (Alper & Holmes, 1983), however in around 1 in 20,000 births a newborn has a naevus of >20 cm diameter when projected to adult size (Castillas et al., 1981; Price & Schaffer, 2010). Based on the size and number of the CMN, individuals may also have a slightly increased risk of developing malignant melanoma within the skin or the Central Nervous System (0.7% overall risk, increasing to 2.5% in large CMN; (Kinsler et al., 2017). CMN can be associated with other complications related to the skin, such as skin fragility, reduced sweating and pruritus (Masnari et al., 2019).

Craniosynostosis: Craniosynostosis has been defined as the premature fusion of the cranial suture which can result in an unusual head shape (D. Johnson & Wilkie, 2011). Craniosynotosis affects between 1 in 1,800 and in 2,000 in the UK (Headlines charity, 2021). Of these, approximately 75% are non-syndromic (involving on cranial suture and not affecting other parts of the skill, face, or body. The prevalence of craniosyntosis is 7.2 per 10,000 live births (Cornelissen et al., 2016).

Ectodermal Dysplasia: Ectodermal dysplasia is a group of closely related conditions which are characterized by changes to the structure of parts of their body which have part of very early fetal development (e.g., the ectoderm: teeth, hair, nails, and sweat glands; Ectodermal Dysplasia Society, 2022). More than 180 different types of Ectodermal Dysplasia have been identified. Depending on the type of condition, Ectodermal Dysplasia can also affect the skin, the eyes or ears, lining of the airways, and the development of fingers and toes, nerves, and other parts of the body.

Eczema: Eczema (or dermatitis) is a non-contagious, inflammatory dry skin condition that can affect people across the lifespan. The most common form of eczema, atopic eczema, affects 1 in 5 children and 1 in 10 adults in the UK (National Eczema Society, 2022). Symptoms of eczema include skin inflammation and dryness, redness, and itching (Odhiambo et al., 2009). Alongside these symptoms ongoing itching can also cause affect other life domains, such as sleep quality (Silverberg et al., 2015). Flare ups of eczema symptoms are often caused by environment factors such as, allergies, clothing, diet, and weather (National Eczema Society, 2022).

Facial Palsy: Facial palsy is a facial visible difference which is caused by weakness of the facial muscles due to temporary or permanent damage to the facial nerve (Hamlet et al., 2021). Bell's Palsy is the most common form of facial palsy, accounting for approximately half of all cases. Bell's Palsy affects around 20.2 people per 100,00 in the UK (Rowlands et al., 2002). Facial palsy can also be caused by trauma to the facial nerve (e.g., surgery or illness) or congenital conditions, such as Moebius Syndrome (Hamlet et al., 2021). Facial palsy can also significantly affect functionality. Verbal and non-verbal communication are often affected, due to lack of mobility in the facial muscles (Keillor et al., 2002). Individuals with facial palsy can also experience dry eyes, drooling, facial spasms and difficulties with eating and drinking (Benecke, 2002).

Limb difference: Limb difference is an umbrella term for conditions or injuries which result in a visible difference to the limbs. This can include congenital conditions which cause differences in the development of the limbs (e.g., congenital hand differences, Reach, 2022) and differences acquired through illness or injuries (e.g., amputation). Limb differences can result in differences in appearance and functionality, due to difficulties with mobility.

Microtia: Microtia is a congenital craniofacial condition which can result in the malformation or misshape of the external structure of the ear (Hamlet & Harcourt, 2020). Approximately 1 in 6,000 people are born with Microtia in the UK. Microtia can occur in isolation or as part of other conditions, such as Treacher Collins syndrome (Zim et al., 2017). This condition can occur alongside atresia, which may require surge to restore hearing.

Vitiligo: Vitiligo is an acquired condition which is characteristic by de-pigmentation of patches of skin (Zhang et al., 2016). These patches can occur on different parts of the body, as well as on mucous membranes. A systematic review suggested that the pooled prevalence of vitiligo across 82 population-based studies was 0.2% of the overall population (Zhang et al., 2016).

Variations in sex development: Variations in sex development is a broad term for a group of condition which cause variation in the development of the internal and external reproductive systems. The most common cause is Congenital Adrenal Hyperplasia (CAH), which results in a lack of the enzyme required to make cortisol and aldosterone (Sanders et al., 2021). This causes the body to produce more androgens and result in differences to the appearance of the genitals (Speiser et al., 2018). CAH affects between one in 10,000 and one in 18,000 children born each year in the UK (GOSH, 2020)

Appendix C Mixed methods quality framework

Stage of study	Domains of quality	Items within domain	Definition of item	Source of domain and items	Application in PhD
Planning	Planning quality	Foundational element	Comprehensible and critical review of the literature is needed to situate the study and shape both the research question and methods	Dellinger and Leech (2007)	An initial literature review was conducted at the beginning of the PhD to establish existing knowledge and identify gaps in research. This formed the basis of the PhD.
		Rational transparency	Justification for using a mixed methods approach is provided	Caracelli & Riggin (1994) Creswell (2003)	MMR meets the aims of the research to develop support materials. The initial study required an exploratory approach. The is a quant investigation in a larger population to confirm.
		Planning transparency	Details should be given about the paradigm, planned design, data collection, analysis and reporting using Creswell's guide to a good proposal	Creswell (2003)	Research paradigm was discussed at length with supervision team and used to guide research design A discussion on paradigm, design and data collection was included in the progression report and discussed during progression review 1 in 2019.
		Feasibility	The design, and each component can be undertaken in the resources available.	(O'Cathain, Murphy & Nicholl (2008)	During PhD registration, a RD1 form was completed and the PhD design is checked for feasibility given the resources. In addition, the PhD timeline is reviewed at regular intervals with supervision team.
Undertaking	Design quality	Design transparency	Description of design type from known typology, or key aspects of design if known typologies do not describe design used.	Creswell & Plano Clark (2007) O'Cathain et al. (2008)	Design described and laid out in detail at the beginning of the PhD as part of PhD registration. All elements of PhD design were aspects of known typology.
		Design suitability	The design is appropriate for addressing the overall research question, matches the reason for combining methods and is appropriate for the stated paradigm	Teddlie & Tashakkori (2009) Creswell & Plano Clrak (2007) Caracelli & Riggin (1994) Onwuegbuzie & Johnson (2006)	Rationale for design identified early in PhD registration and development process. MMR to meet pragmatic aim of the development of evidence base for support materials.
		Design strength	The strengths and weaknesses of methods are considered to minimise shared bias and optimise the breadth and depth of the study	Caracelli & Riggin (1994) Onwuegbuzie & Johnson (2006)	Chosen designs evaluated during the PhD design and continually throughout the PhD, at progression milestones and with supervisory team. Evaluation is also incorporated into thesis chapters.

		Design rigour	Methods are implemented in a way that remains true to the design	Creswell & Plano Clark (2007) Caracelli & Riggin (1994)	As above, rigour was reviewed and discussed regularly with supervisory team and at progression milestones. Reflections on this are also incorporated into thesis chapters.
Interpreting	Interpretive rigour (Conclusions are based on the findings)	Interpretive transparency	It is clear which findings have emerged from which methods	O'Cathain et al. (2008)	The mixed methods research has been conducted using a co-ordinated (rather than integrated design) so data was collected from methods separately and then integrated at a later point.
		Interpretive consistency	Inferences are consistent with the findings on which they are based.	Teddlie & Tashakkori (2009)	A thorough process of checking data against themes has been undertaken and themes have been revised in line with this (qualitative data). A similar rigorous process was applied to ensure that statistical analyses on quantitative data are appropriately interpreted.
		Theoretical consistency	Inferences are consistent with current knowledge or theory.	Teddlie & Tashakkori (2009) Dellinger & Leech (2007)	The inferences drawn from data are line with current theory. E.g., Template analysis was used which incorporated some pre-existing constructs from literature and theory. This was also examined during the writing of the thesis chapter for the first study. A similar process was undertaken with findings from later studies.
		Interpretive agreement	Others are likely to make reach the same conclusions based on the findings presented, including other researchers and participants.	Teddlie & Tashakkori (2009) Onwuegbuzie & Johnson (2006)	For the qualitative data collected, peer debriefing has been undertaken with the supervision team. A second coder also reviewed a broad selection of the transcript.
		Interpretive distinctiveness	Conclusions drawn are more credible than any other conclusions.	Teddlie & Tashakkori (2009)	Conclusions drawn from the studies of the PhD have been discussed at length with PhD supervision team. This has been necessary as the mixed methods findings informed the development of the intervention. It was therefore essential to ensure

					that the conclusions of the first study were sound and grounded in the data.
		Interpretive efficacy	Meta-inferences from the whole study adequately incorporate inferences from the qualitative and quantitative findings and inferences.	Teddlie & Tashakkori (2009) Onwuegbuzie & Johnson (2006) O'Cathain et al. (2008)	Integration of quantitative and qualitative was discussed and reviewed in depth with the PhD supervision team. It was essential to clearly demonstrate the process and outcome of this integrate within the thesis. The meta-inferences also informed the final stage of the PhD so it will be important to ensure that the conclusions drawn are sound and grounded in the data.
		Interpretive bias reduction	Explanations are given for inconsistencies between findings and inferences.	Caracelli & Riggin (1994) Creswell & Plano Clark (2007) Teddlie & Tashakkori (2009)	The literature on managing discrepancies in data has been reviewed and a plan for how to work with this data was developed with the supervision team. Any conflicting data within the qualitative data was reported in the thesis to demonstrate the range of experience of parents.
		Interpretive correspondence	Inferences correspond to the purpose of the study, the overall research question and the research questions within this.	Teddlie & Tashakkori (2009)	The researcher frequently referred back to the aims, objectives, and research questions throughout all stages of the PhD. The research aim has informed the research paradigm as well as design so is integral to the project. This is a process that is facilitated by holding the task of writing the PhD thesis in mind, to ensure that the overall story of the PhD is in line with aims and research questions.
Interpreting	Inference transferability	Ecological transferability	Transferability to other contexts and settings	Tasakkori & Teddlie (2003. 2008. 2009)	The second study of the PhD aimed to sample from outside of the usual research centre methods e.g., using Reddit.
		Population transferability	Transferability to other groups and individuals		Strategies to increase generalisability have been utilised such as the development of a purposive

					sampling framework based on theory and literature of visible difference and body image.
		Temporal transferability	Transferability to the future		The PhD aimed to develop an evidence base to inform future support for parents, therefore has a focus on how the research might be utilised in the future. The support provision has changed over the period of this PhD, and this continue to be reviewed and considered.
		Theoretical transferability	Transferability to other methods of measuring behaviour		Quite a number of methods have been used over the course of this PhD (focus groups, interviews, surveys). Each method is described in detail so that a reader can judge the transferability of the findings.
Disseminating	Reporting quality	Report availability	Study is successfully completed within allocated resources of time, money and staff	Dalta (1997)	At PhD registration, the proposed studies were assessed to determine whether the studies could be done on time given the resources. This timeline was reviewed at regular intervals and adapted to ensure that the PhD will be completed in the allocated time.
		Reporting transparency	Key aspects of study reported, according to GRAMMS	Caracelli & Riggin (1994) Creswell & Plano Clark (2007) O'Cathain et al. (2008)	All aspects of the research are described in depth when including in the write up of the PhD thesis.
		Yield	Whole more than the sum of the parts	O'Cathain et al (2007)	This was carefully considered during the integration phase and discussed at length with the supervision team. A detailed plan was developed to interpret and integrate the findings from both quantitative and qualitative elements.

Application in the real world	Synthesisability (Of sufficient quality for inclusion in systematic reviews)	12 quality criteria: 6 for qualitative research 3 for quantitative experimental 3 for quantitative observations 3 mixed methods	An example criterion is: “justification of the mixed methods design”	Pluye, Gagnon, Griffiths & Johnson-Lafleur (2009)	Criteria utilised for systematic reviews was mapped onto research design (as far as possible).
	Utility	Utility quality	The findings are used by consumers and policy makers	Caracelli & Riggin (1994) Dalta (1997) Dellinger & Leech (2007) Onwuegbuzie & Johnson (2006) Dellinger & Leech (2007)	A main aim of the PhD was to develop an evidence base for the development of support materials for parents. Therefore, it is hoped that the findings will have real world applications through the development of support materials which can be used by parents and professionals who support them.

Appendix D: Impact of Public Involvement

Public and Patient Involvement (PPI) activity	Feedback	Changes made/action taken
Public involvement focus groups at the AC parent support workshop: Focus group activity consisted of professionals carrying out a priority setting task for future parent support	Professionals prioritise: Content of support: • Support for parents managing emotions • Managing others' reactions • Managing relationships • Peer support • Decision making Mode of delivery: • Across the lifespan • Accessibility • Education and awareness raising • Normalising support • Being mindful of language	Record of discussion taken for later review and reflection. Feedback from this activity is anticipated to be particularly useful for considering variables for study 2 and intervention development. Reflected upon feedback during study 1 analysis when developing coding template and during coding.
Members of parent advisory group (4 parents) provided feedback of study 1 research materials	• Using more condition specific language for each charity approached in order to better resonate with parents • More examples of visible differences on materials so parents understand what kind of conditions might be included • Add a question in the interview about age of child affected • Make parent advert more colourful • Be flexible with interview schedule, let parents tell their story naturally	• Language in materials personalised to suit each charity and corresponding parent populations • Increased number and variety of examples of visible differences on information sheet • Age of child asked in Qualtrics form but also confirmed with parent at the beginning of the interview • Altered parent advert to make more colourful • Spoke with parent about the nature of semi-structured interviews and how they are able to be flexible and asked for her thoughts on whether this was appropriate in relation to this feedback

Parent of child with and charity director of Caring Matters Now provided feedback on focus group materials	• Small changes in language e.g., change family to carers to reflect the people who might be attending the event	• Changes made to language in information sheet as suggested
Members of parent advisory group (4 parents) received a summary of the findings of study 1 and provided feedback on conclusions and next steps.	• The findings resonated with their own experiences of caring for their child • Small errors within the research summary • Would be helpful to disseminate to members through emails and blog posts	• Correction of errors • Dissemination through blog posts on charity websites • Dissemination to participants through email
Members of parent advisory group (3 parents) provided feedback on study 2 research materials and recruitment plan.	• Good level of information provided at the beginning of survey • Design and format of survey is engaging • Estimation of time to complete between 15 and 25 mins • Small errors within survey e.g., typos and grammar • Likert scale options could be made clearer • Positive feedback about the addition of COVID-19 open-ended questions	• Correction of typos • Edit headings on Likert scale for ease of comprehension • Include open-ended questions on experience of COVID-19 • Member of the parent advisory group supported in the creation of a recruitment video for study 2.
Academic in the visible difference field who is also a parent reviewed the study 2 survey materials and provided feedback on recruitment plan.	• Suggestions to focus recruitment efforts during the school holidays as a time of relative normality after several school terms affected by COVID-19 • Some issues with skip/display logic on SPSS	• Plan recruitment to begin in the second week of school holiday. • Correct errors in skip/display logic.
Members of parent advisory group (3 parents) received a summary of the findings of study 2 and provided feedback on conclusions and next steps.	• Interesting results which make sense in the context their own experience • Some concepts may need clarification or further explanation (e.g., psychological flexibility) • Expressions of interest in being involved in intervention development	• Add additional detail to explain psychological concepts • Invitation to take part in PAR stages of intervention development – includes review of mixed method conclusions

Participatory Action Research approach to intervention development	See chapter 8	See chapter 8
Members of the parent advisory group provided feedback on the materials for the PAR workshops.	• Feedback on subscales of eHIQ • Change in language from "intervention materials" to "parenting toolkit"	• Third subscale not included as items did not feel relevant • Change language in survey to increase accessibility.

Appendix E: Reflection on the impact of COVID-19

At the beginning of 2020, the world was hit by a novel virus SARS CoV-2-virus (COVID-19). On March the 23rd 2020 the UK government implemented policies to slow the spread of the virus. At this point in the PhD, data collection was about to begin for study 2. Adaptations had to be made to the PhD timeline and methods to adjust to the new situation.

Firstly, a primary outcome of the second study of the PhD was parental negative affect. Due to COVID-19 restrictions (e.g., home schooling) and general concerns about health and infection, parental anxiety and general negative affect were likely elevated during this time. As such, collecting data during the COVID-19 outbreak may have caused bias in the data. This would be particularly detrimental because the integrated findings of the studies 1 and 2 informed the intervention development. Therefore, it was imperative to reduce the impact of confounding variables by delaying data collection.

Some COVID-19 restrictions directly affected some aspects of living with a visible difference. As discussed throughout the thesis, many young people with a visible difference experience appearance-related social challenges, such as teasing or unwanted attention (Rumsey & Harcourt, 2007). During lockdown it was only possible to socialise within households and with friends and family on virtual platforms. This meant young people with visible differences were less likely to interact with people outside their immediate social circle and might experience fewer social challenges. Consequently, parents' responses to questions about social challenges may not be typical during this time. On the other hand, increased use of video calling software might be challenging for young people with visible facial differences. This could cause young people to notice their visible difference more frequently and cause greater appearance-related distress. Observing this distress may also be challenging for parents. Parents may also have been concerned about how their child may cope in the future, after lockdown restrictions end and their child returns to social or public settings.

In addition, many children with a visible difference also have underlying health conditions. An underlying health condition may be the cause of a child's visible difference (e.g., ectodermal dysplasia, alopecia), or a consequence of the condition/injury (e.g., burn injury) or related treatment (e.g., immunosuppressant treatment for conditions such as psoriasis). Some underlying health conditions may result in children being more vulnerable to COVID-19 (Williamson et al., 2020). For example, children with ectodermal dysplasia can experience cardiomyopathy (a disease of the heart muscle which affects circulation of blood). It has been suggested that cardiomyopathy may put individuals at increased risk of developing severe illness if they contract COVID-19 (Cardiomyopathy UK, 2020). Consequently, parents of children with an underlying health condition alongside their visible difference, may have experienced heightened anxiety during the pandemic. Due to increased pressure on health services, parents may also have been concerned about the impact of the pandemic on their child's present or future treatment (e.g., cancelled appointments, delays in surgery). As discussed above, this increased anxiety in parents could have been a possible confounding variable in the data collected for study 2.

As a result of these factors, I took the decision to suspend my PhD studies for a period of 3 months. When my PhD recommenced, I made several changes to the design and recruitment plan for the second study and future studies. Firstly, all recruitment and data collection moved online. This was not a huge shift from the original design of the second study which was a survey hosted on the online platform Qualtrics. However, recruitment at in-person charity events and providing the option of hard copies of the survey had been included in the original recruitment and data collection strategy. This approach may have helped to recruit a more diverse sample by reaching those who may be less likely to complete an online survey (see section 3.10.3. on digital exclusion). Furthermore, recruitment through events in 2018-2019 had been successful in recruiting more fathers to the PhD, as they had attended with their partners. This kind of large-scale snowball sampling was more difficult to achieve once recruitment moved entirely online.

A specific change was also made to the design of study 2. Open-ended questions asking parents about their experiences of the COVID-19 pandemic and lockdown were added to the end of the online survey. Recent research with parents of children with CLP has found that the changes to healthcare and daily functioning during the pandemic has resulted in emotional distress for caregivers (Costa, McWilliams, et al., 2021). Consequently, it was deemed important to provide parents with an opportunity to share their experiences of the pandemic in their own words. This qualitative data also provided context to the answers given to quantitative measures (e.g., less reported social challenges due to less social contact).

Completing a PhD during a global pandemic has been difficult. Managing personal loss and health concerns, alongside anxiety about the broader impact of this crisis on the world has been incredibly anxiety inducing and adapting a PhD to fit into the new world has presented a range of challenges. However, the new way of working more autonomously, whilst physically and emotionally isolating at times, created space for me to become more confident and independent in my abilities and decisions. Although this is a process all post-graduate researchers experience, it felt somewhat accelerated to me due to the necessity of home working caused by COVID-19. Despite the challenges outlined, the PhD program of work was also able to continue and produce new knowledge and understanding to the field of visible difference.

Appendix F: Study one materials

Parent interview schedule

Demographic questions

What is your age?

How would you describe your ethnic background?

What is your marital status?

Who do you usually live with?

Do you have a religion?

What's your highest level of qualification?

Are you currently working?

What job do you/ did you do?

Family context

How many children do you have?

How old is your child?

Can you tell me about your child's condition and how it affect their appearance?

Can you tell me about any treatment your child has received for their condition?

Parenting experiences

As a parent, what has your experience of your child's condition/injury been like so far?

Prompts:

Have there been any particular times when things have been more challenging?

Diagnosis/injury, treatment, decision making, school, social experiences, future concerns

If you have other children…what is their relationship like with their sibling? / How do you think your child's condition or injury affects their sibling?

Can you tell me about any positive experiences that you may have had as a result of your child having an appearance altering condition/injury?

Parent experience – coping

How do you feel about your child’s visible difference when you are with your family/ extended family/ friends/ out in public?

What things in your life have helped you to cope with challenges related to your child’s condition/injury?

Prompts:

Social support (partner support, family support, friend support), confidence, resilience, optimism, attitudes about appearance, coping strategies

Support experiences

Can you tell me about any support that you have received as a parent?

Prompts:

Thinking about the experiences you have just spoken about can you tell me what have been the good experiences and what you feel could have been better?

What has made it easier/harder for you to access support?

Who has that support been from?

Thinking about the support that you have had; can you describe any times when you feel that more support would have been beneficial?

And what additional support you would have liked?

Prompts:

For you as a family, how would that have been beneficial?

How have your thoughts about what support you might want/might have wanted changed over time?

How would you have liked that support to be delivered?

Thinking ahead to the future, are there any specific things you anticipate that you would want support with as a parent?

Prompts:

For you as a family, how would that support be beneficial?

In what format do you think it would be helpful to deliver that kind of support in?

Professionals interview schedule

Demographic questions

Can I ask how old you are?

How would you describe your ethnic background?

What is your marital status?

Who do you usually live with?

Do you have a religion?

What’s your highest level of qualification?

What is your job title?

Can you describe what your role in supporting families affected by an appearance altering condition/injury?

Can you describe for me what you feel the support needs of parents with a child with an appearance-altering condition/injury are?

Prompts:

What concerns/problems/challenges are coming to you with?

What are the key time points for support on the patient/service user journey?

Can you describe any factors that you feel contribute to helping parents to adjust well to their child’s condition/injury?

Prompts:

Social support (partner support, family support, friend support), confidence, resilience, optimism, attitudes about appearance, coping strategies

What interventions/support do you provide/ are you aware of for parents of children with an appearance altering condition/injury?

Prompts:

Give examples e.g., online information, leaflets, groups etc.

What do you think/in your experience may be some of the barriers to delivering this support to parents/parents accessing this support?

What do you think should be included in support materials addressing common challenges for parents of children with an appearance altering condition/injury?

Prompt:

What challenges do you think support materials aim to address?

What format should support materials be delivered in?

Focus group protocol

CMN focus group protocol for co-facilitators

Top Tips:

- **Try and involve everyone in the group** – prompt those who may not be speaking with questions e.g. Did you feel the same way? What do you think about this? (This can happen particularly when there are couples present and one person speaks for both, we want to avoid this)
- **Take notes -** particularly of interesting points that you might want to ask follow-up questions on, so that you don't forget these points even if conversation moves on a bit
- **Keep parents focussed on how THEY feel** – because these parents will be so focussed on their child, they can quite easily slip into how their child is feeling and managing. Try and keep them focussed with questions like, and what is was/is that like for you as a parent?, when that happened how did it make you feel?
- **Parents might get upset –** if this happens just ask them quietly if they're ok, remind them that they don't have to continue, ask them if they would like to take a break and remind them that they can leave at any point (I will also have tissues on hand)
- **Parents may be quite negative about healthcare and support services –** try not get too involved in this but have an empathetic response e.g., "that sounds very difficult for you", however do feel free to ask follow-up questions to better understand their experience
- **Parents may ask you advice on how to handle things** – if this happens be sympathetic and validate that what they are experiencing sounds challenging and direct them to me (I might know an appropriate that I might have come across) OR good general advice is signposting them back to CMN resources (staff available at the event, website), Changing Faces or suggest they ask their healthcare teams
- **Keep an eye on the recorder** – I will do checks before we start on battery life and memory but just check on them every now and then as it is quite a long session, just to make sure they are still recording
- **Small children might accompany their parents** – this often happens with parent focus groups even if there are other activities happening, it does make conducting the focus group a little harder, but we just need to be flexible around this

Things that I anticipate will come up (can also use as prompts if things get a bit quiet):

- Psychological distress (parents managing their own reactions)
- Concerns about the future
- Social situations (e.g., unwanted attention in public, difficulties interacting at school)
- Dealing with health professionals
- Managing the responses of others (e.g., other family members)

- Support seeking behaviours (e.g., information seeking)
- Support from other people (e.g., friends, family, peer support)

How the session will run:

Parents/carers will arrive and be asked if they have read the information sheet on the door and be asked to sign a consent form (there will be spare information sheets on the door for those who haven't had a chance).

I plan to try and organise families by age of their child, so when they go in, I will ask them how old their child is and assign them a group (we may have to move people around if the numbers in groups don't quite work).

There will be a form on each table to record demographic information for each group, please encourage parents to fill it out as they sit down and then we can always try and fill in any gaps as the session goes on. There will also be sticky labels for name tags.

Once everyone is in and sat down, I'll introduce us and give a quick bit of context to the session and ask if everyone has had a chance to read the information sheet and sign the consent form. I will give you both spare copies in case someone in your group hasn't.

The session is an hour and a half long and will have 4 activities (+ an introduction activity):

- **Introduction (5 mins):**

In their groups I will ask parents to go around and say their name (also encourage name tags for your benefit and to help conversation between parents), how old their child is and where they are from

- **Activity 1: Challenges experienced (Equipment: challenges flipchart sheet, pens, your phone)**

20 mins

I will introduce the activity as a chance for parents to think and reflect on the challenges that they have faced as a family so far and share these with the group and make a mind map using the flipchart paper.

You will each be with one group for this activity. Your role will be to listen to the discussion and make some notes about what you hear.

The kinds of notes that would be helpful would-be general themes that emerge from the conversation. I am anticipating that the kind of things that will come up are parents coping emotionally, difficulty with social situations, school, challenges with health professionals, worries about the future, how to talk to their child etc. If you have time and you hear particular examples that you think are interesting, please jot these down too.

It might be helpful to answer follow-up or probe questions during this discussion if you want to seek clarification or hear more about an interesting experience e.g., can you say a bit more about that? Can you explain what you mean by….? Why do you think that was? Why do you think that was particularly challenging?

Although some follow-up questions may be useful you don't need to facilitate the whole conversation, parents will want to talk to each other so just let the conversation flow naturally, UNLESS it gets really off topic then gently guide them back to the activity, maybe with a question or comment about something someone has said.

With 5 mins to go I will ask the groups to summarise their discussions to you (the facilitator). This will help me to analyse when I go back and listen to the recording, I am essentially asking them to do a very quick thematic analysis and pull out the main themes of their conversations.

At the end of the activity please take a photo of the flipchart paper and collect it in.

- **Activity 2: Social situations (Equipment: social situations flipchart sheet, pens, post-its, your phone)**

10 - 15 mins

I will introduce the activity as an exercise in thinking about their family and their child in social situations and how comfortable they are. I will ask parents to use their post-it notes to place on the social situation circles and write on the notes how comfortable they feel in each of these situations on a scale of 0 (very uncomfortable, would avoid) to 10 (completely confident) on the post-its. I will also encourage them to talk about experiences they might have had good and bad in different social situations as they do this.

There will also be pens so parents can add their own social situations to the flipchart paper if I have missed anything out

As they go along, please make notes of their discussion. If parents are just putting on post-it notes and not discussing, please can you ask them questions about their experiences and encourage them to explore it a bit.

Again, at the end of the activity please take a photo of the flipchart and collect it in.

- **Activity 3: Life stages activities (Equipment: life stages flipchart sheet, pens, post-its, your phone)**

10 - 15 mins

I will introduce the activity as an exercise in thinking about different life stages for their child and what either their experiences have been/ what they are worried about in the future. Will ask the parents to use post-it notes to write their experiences/worries and then place them on the life stages points on the flipchart paper.

There will be pens so parents can add extra life stages to the flipchart if I have missed anything out.

Again, as they go along, please make notes of their discussion. If parents are just putting on post-it notes and not discussing, please can you ask them questions about their experiences and encourage them to explore it a bit.

Again, at the end of the activity please take a photo of the flipchart and collect it in.

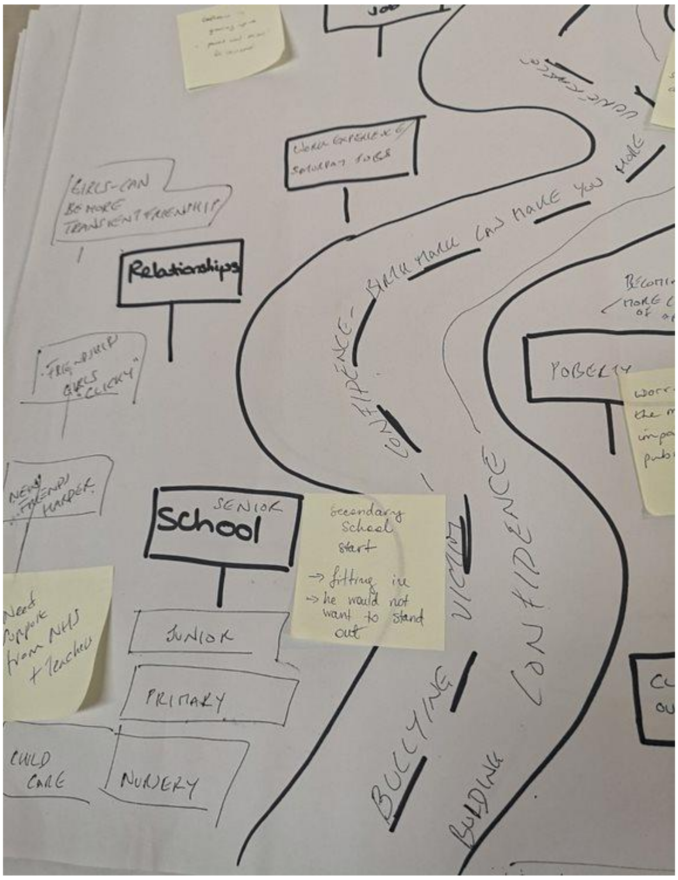
- **Activity 4: Support experiences (Equipment: support experiences flipchart sheet, pens, your phone)**

10 mins

I will introduce the activity as an opportunity to talk about support experiences that they have had good and bad and/or support they would like to have had/would like to have in the future.

Again, please take notes as in previous activities and take a photo of the flipchart and collect it in at the end.

Examples of focus group visual aids



Study 1 template analysis coding templates

Parent interview final coding template

1. **Impact on parent wellbeing**
    - 1.1. Guilt
        - 1.1.1. I caused this
        - 1.1.2. Decision making
    - 1.2. Low mood
    - 1.3. Anxiety
        - 1.3.1. The Unknown
    - 1.4. Initial reactions
        - 1.4.1. The initial shock
            - 1.4.1.1. This is not what I expected
        - 1.4.2. Isolation
        - 1.4.3. Learning over time
    - 1.5. Practical challenges
        - 1.5.1. Burden of care

2. **Child's reaction**
    - 2.1. Child resilience
        - 2.1.1. Child not concerned
        - 2.1.2. Social coping
    - 2.2. Individual differences
    - 2.3. Changes with age
    - 2.4. Siblings

3. **Navigating the reactions of others**
    - 3.1. General public
        - 3.1.1. Questions and comments
        - 3.1.2. Unsolicited advice
        - 3.1.3. Adults are worse
            - 3.1.3.1. Other parents
    - 3.2. Family conflict
        - 3.2.1. Grandparents
        - 3.2.2. Extended family

4. **Parent-child relationship**
    - 4.1. Coping link
        - 4.1.1. Child distress is parent distress
    - 4.2. Parent attachment
        - 4.2.1. Difficulty with bonding
    - 4.3. Parent-child communication
        - 4.3.1. Child modelling
        - 4.3.2. Talking about difference

        - 4.3.2.1. Child narrative
        - 4.3.2.2. Talk about diversity
    - 4.3.3. Shared decision making

5. **Parent appearance attitudes**
    - 5.1. Gender differences
        - 5.1.1. Worse for girls
        - 5.1.2. Functional concerns in boys
    - 5.2. More than appearance
        - 5.2.1. Appearance altering treatment not important
        - 5.2.2. Appearance secondary to function
        - 5.2.3. What is normal?
            - 5.2.3.1. It’s normal to us

6. **Protect my child**
    - 6.1. Being strong for the child
    - 6.2. Future concerns
        - 6.2.1. Bullying and social exclusion
        - 6.2.2. Transitions
            - 6.2.2.1. Moving and starting school
            - 6.2.2.2. Teen years
            - 6.2.2.3. Relationships and dating
            - 6.2.2.4. Work
        - 6.2.3. Child mental health
        - 6.2.4. Body image concerns
    - 6.3. Concealing difference
        - 6.3.1. Strategies to conceal
        - 6.3.2. Conflict about concealment
        - 6.3.3. No need to hide

7. **Existing support**
    - 7.1. Being heard
        - 7.1.1. Charity support
        - 7.1.2. Specialist healthcare professionals
    - 7.2. Friend and family support
        - 7.2.1. Partner support
        - 7.2.2. Sibling relationships
        - 7.2.3. Family history
    - 7.3. Accessible support
        - 7.3.1. Geographic barriers
        - 7.3.2. Time barriers
        - 7.3.3. Finding support
    - 7.4. Education
        - 7.4.1. Information seeking
            - 7.4.1.1. Doctor Google
        - 7.4.2. Raising awareness
    - 7.5. Double edged sword

        - 7.5.1. Health professional experiences
        - 7.5.2. Social media
    - 7.6. Gaps in support

Parent focus group coding template

1. **Impact on parent wellbeing**
    - 1.1. Guilt
    - 1.2. Anxiety
        - 1.2.1. The unknown
        - 1.2.2. Fear
    - 1.3. This is not what I expected

2. **Navigating the reactions of others**
    - 2.1. General public
        - 2.1.1. Questions and comments
        - 2.1.2. Unsolicited advice
        - 2.1.3. Adults are worse
    - 2.2. Family and friends
        - 2.2.1. Other generations
        - 2.2.2. Siblings

3. **Parent-child relationship**
    - 3.1. Parent-child communication
        - 3.1.1. Child narrative
        - 3.1.2. Shared decision making
    - 3.2. Child modelling
    - 3.3. Child led

4. **Protect my child**
    - 4.1. Future concerns
        - 4.1.1. Bullying and social exclusion
        - 4.1.2. Child mental health
        - 4.1.3. Body image concerns
        - 4.1.4. Transitions
            - 4.1.4.1. Moving and starting schools
            - 4.1.4.2. Relationships and dating
            - 4.1.4.3. Work
    - 4.2. Concealing difference
        - 4.2.1. Embrace not hide
        - 4.2.2. I would change it
        - 4.2.3. Appearance altering treatment not worth it

5. **Existing support**
    - 5.1. Being heard
        - 5.1.1. Family and friend support
        - 5.1.2. Specialist professionals

        - 5.1.3. Reducing isolation
    - 5.2. Health care challenges
        - 5.2.1. Health professional communication
        - 5.2.2. Lack of awareness
        - 5.2.3. Rare condition, health professional interest
    - 5.3. Education
        - 5.3.1. Information seeking
        - 5.3.2. Awareness raising
        - 5.3.3. Doctor Google
    - 5.4. Gaps in support

Health professional interviews

1. **Impact on parent wellbeing**
    - 1.1. Negative psychological impact
        - 1.1.1. Guilt
        - 1.1.2. Feelings of failure
        - 1.1.3. This is not what I imagined
    - 1.2. Individual coping style
    - 1.3. Practical challenges
        - 1.3.1. Financial
        - 1.3.2. Sleep
        - 1.3.3. Burden of care
            - 1.3.3.1. Own needs are second

2. **Navigating the reactions of others**
    - 2.1. General public
        - 2.1.1. Questions and comments
    - 2.2. Friends and family
        - 2.2.1. Conflict within families
        - 2.2.2. Partners
        - 2.2.3. Siblings

3. **Parent-child relationships**
    - 3.1. Parent-child attachment
        - 3.1.1. Parent-child bonding
    - 3.2. Parent-child interaction
        - 3.2.1. Child narrative
        - 3.2.2. Child modelling
        - 3.2.3. Child involvement in decision making

4. **Appearance attitudes**
    - 4.1. Appearance is important
        - 4.1.1. Appearance altering treatment
    - 4.2. Minimising difficulties
        - 4.2.1. It’s not JUST appearance

5. **Protect my child**
    - 5.1. Being strong for the child
    - 5.2. Future concerns
        - 5.2.1. Bullying and social exclusion
        - 5.2.2. Child mental health
        - 5.2.3. Body image concerns
        - 5.2.4. Decision making
        - 5.2.5. Transitions
            - 5.2.5.1. Moving or starting schools

6. **Existing support**
    - 6.1. Health professional communication
        - 6.1.1. Language
        - 6.1.2. Misinformation
        - 6.1.3. Stigma
    - 6.2. Out of our depth
        - 6.2.1. Gap in psychological services
        - 6.2.2. Lack of awareness

Appendix G: Mapping qualitative themes to psychosocial constructs

Corresponding theme(s) from study 1	Construct and rationale	Possible measures	Items	Psychometrics and other info
“It broke me”	**A. Parent Distress** (Anxiety and Depression/Low mood experienced by parents) **Rationale for inclusion:** **Evidence from study 1:** Study 1 found that parents experience anxiety and low mood both when their child is initially diagnosed/injured and in some this can also persist long-term. This can be related to challenges that emerge as the child grows up e.g. worries/feeling low when anticipating threats to child, worries about arming the child to manage challenge, feeling low when parent feels they have not “got it right” for their child **Support from the literature:** The existing literature also reports that parents experienced distress • Distress at diagnosis or injury (Gasser, -Bellet, & Hohlfeld, 2004; N. M. Stock & Rumsey, 2015; Heath et al. 2018)	**1. Hospital Anxiety and Depression Scale (HADS)** Zigmond and Snaith (1983) (Widely used in vis dif literature and vis dif parent literature)	14 items in 2 subscales, one for anxiety and one for depression Rated on 0-3 scale of not at all to most of the time How you have been feeling in the last week: A: I get tense or wound up I get a sort of frightened feelings as if something awful is about to happen Worrying thoughts go through my mind I can sit at ease and feel relaxed I get a sort of frightened feeling like ‘butterflies’ in the stomach I feel restless as I have to be on the move I get sudden feelings of panic D: I still enjoy the things I used to enjoy I can laugh and see the funny side of things I feel cheerful I feel as if I am slowed down I have lost interest in my appearance I look forward with enjoyment to things I can enjoy a good book or radio or TV program	“Good” reliability and validity Zigmond and Snaith (1983) Moss et al (2014) anxiety alpha= .83, depression alpha= .82 Bjelland et al (2002) 6 studies reported correlations between BDI and HADS-D .62 and .73, HADS-A .61 to .83 and HADS total score Two studies found correlations between GHQ-28 and HADS-A .50 and .68, correlations between the clinical anxiety scale and HADS-A .69 and .75 Used in a number of vis dif parent papers, Nelson et al (2009) causal attributions paper, Stock et al (2019) risk and protective factors for CLP

	• Guilt (Hawkins et al., 2018; Nelson et al., 2009; Stock and Rumsey, 2015) • Psychological isolation (Heath et al, 2018; Tanner et al., 1998) • Anxiety and concerns social stigma (Hlongwa & Rispel, 2018, Klein et al., 2006 Rumsey & Harcourt, 2005)	2. **Positive and Negative Affect Scale (PANAS)** Watson et al. (1988)	20 items 2 10 item subscales Indicate the extent you have felt this way over the past week: Positive Affect: Interested Excited Strong Enthusiastic Proud Alert Inspired Determined Attentive Active Negative Affect: Distressed Upset Guilty Scared Hostile Irritable Ashamed Nervous Jittery Afraid	Watson (1988) Positive affect alpha > 0.86 Negative affect alpha > 0.84 Test retest over 8 weeks: PA: r = 0.47-0.68 NA: r = 0.39-0.71 Used in wider paediatric literature: Hexem et al (2013) Emotional expression in parents of children with serious illness (children in palliative care)

“It broke me”	**B. Parenting Stress** (Reactive response of parents to challenges, in this case arising from child’s visible difference. Where the threat outweighs parents perceptions of their ability to cope)	1. **Parenting Inventory for parents** (Streisand et al., 2001)	Brief PIP Frequency and Difficulty parenting stress 4 subscales: Communication, medical care, Role function, Emotional Functioning Illness specific Communication Waiting for my child’s medical results Talking to my child about their illness Talking to my family members about my child’s illness Medical Care Taking charge of changes to my child’s daily treatment Helping my child with medical procedures	Full scale: Streisand et al. (2001) PIP-F alpha=.96, PIP-D, alpha=0.95 Correlated with measure state anxiety and other measures of parenting stress Brief PIP:

			Accompanying my child during medical tests and treatment Role function: Having little time to attend to my own needs Feeling uncertain about disciplining my child Noticing a change in my relationship with my partner Emotional functioning: Worrying about the long-term consequences of the disease Feeling helpless regarding my child’s situation Feeling uncertainty about the future	Stress of family carers paediatric patients (e.g. asthma, cystic fibrosis) Casana-Granell et al. (2018) Communication alphas F = 0.66, D=0.64 Medical care alphas F = 0.69, D = 0.54 Role function alphas F = 0.42, D = 0.63 Emotional functioning F = 0.77, D = 0.66
		2. Perceived Stress Scales (PSS-10) Cohen et al. (1983)	10 items In the last month… How often have you been upset because of something that happened unexpectedly? How often have you felt that you were unable to control the important things in your life? How often have you felt nervous and “stressed”? How often have you felt confident about your ability to handle your personal problems? How often have you felt that things were going your way? How often have you found that you could not cope with all the things that you had to do? How often have you been able to control irritations in your life? How often have you felt that you were on top of things? How often have you been angered because of things that were outside of your control? How often have you felt difficulties were piling up so high that you could not overcome them?	Nelson et al causal attributions paper, used by Stock et al (2019) in CLP parent’s risk and protective factors Hemati et al – CLP Iranian study Associated with similar constructs and health constructs Cobb et al 1991 In burn injured patient’s alpha = all greater than .84 Test-rest of 2 days 0.85

Being “battle” ready	**C. Social confidence in parents** (Parent confidence in social situations relating to their child’s visible difference e.g., addressing comments from members of the public) **Rationale for inclusion:** **Evidence from study 1:** Parents are concerned with their child modelling their own behaviour when managing social interactions relating to their visible difference e.g., questions and comments from the general public. The data from study 1 indicates that parents who manage these interactions confidently tend to have children who are more confidence and the parent’s themselves also seem less distressed **Support from theory and literature:** • Social Learning Theory (Bandura, 1977) – children model and imitate parent behaviour • SRs found that social interaction skills training are common in psychosocial interventions available for individuals with vis diff (Jenkinson et al., 2015; Harcourt et al., 2018) • SIST found to be effective in reducing parent-reported teasing (Madern et al., 2006), increased self-esteem in cancer patients (Varni et al., 1993), a sig effect on overall communication with peers (Kapp-Simon et al., 2005) and reduced self-report behaviour	**1. CARe Burn scale** Social situations subscale Griffiths et al. (2019)	3 items 1. I feel ok when other people look at my child’s burn wounds/scars 2. I feel ok when other people ask about my child’s burn wounds/scars 3. I feel ok talking to other people about how my child’s burn injury happened Adapted items: 1. I feel ok when other people look at my child’s appearance-affecting conditions or injury 2. I feel ok when other people ask about my child’s appearance-affecting condition or injury 3. I feel ok talking to other people about my child’s appearance-affecting condition or injury	All Scales Cronbach’s alphas > .80 Item total coefficients > .70 Correlates with other validated quality of life questionnaire, particularly social scales of other Burn QOL scales (BSHS)

	problems in burn injuries patients (Blakeney et al., 2005)			
“It broke me” **Appearance does(n’t) matter** **Being “battle” ready**	**D. Psychological flexibility** (The ability for parents to attend to cognitions and emotions with openness and awareness and respond with flexibility to the needs of the situation in line with their values) **Rationale for inclusion:** **Evidence from study 1:** Parents who demonstrate psychological flexibility may be aware of their child’s visible difference and challenges it might pose in an appearance-focused society but strive to set a good model of coping by not avoiding or control challenging situations and communicating openly about the visible difference. This aligns with their desire for their child to adjust well to their visible difference and to prevent these challenges from affecting their child’s wellbeing. **Evidence from the literature:** • In women affected by lipoedema (an acquired visible difference): self-reported psychological flexibility positively predicted quality of life (Dudek et al. 2016) • In parents of adolescents with chronic pain psychological flexibility was significantly negatively correlated with encouraging parent responses	**1. Parenting-Specific Psychological Flexibility (PSPF) Scale** Parent et al. (2015) Brassel et al (2016) Parent specific adaptation of AQA-II	My painful experiences and memories make it difficult for me to parent the way I would value. In my role as a parent, I’m afraid of my feelings. In my role as a parent, I worry about not being able to control my worries and feelings. My painful memories prevent me from having a fulfilling life as a parent. Emotions cause problems in my parenting. It seems like most parents are handling their role as a parent better than I am. Worries get in the way of my success as a parent	Brassell et al. (2016) Sample of children between the ages 3 and 17 Cronbach’s alpha=0.94 Two-week test rest was 0.74 Scale created by adapting the AAQ-II for parenting role.

	(McCracken & Gauntlett-Gilbert, 2011)			
"It broke me" **Walking the tightrope**	**Self-compassion** (Parents ability to practice self-compassion towards themselves when experiencing struggling with challenges related to their child's visible difference) **Rationale for inclusion:** **Evidence from study 1:** Parents who are invested in their role in protecting their child from future and are harsh in the expectations of themselves in this role, experienced distress. Parents who blamed themselves for their child's visible difference also experienced distress **Evidence from the literature:** • In parents of burn-injured children higher self-compassion scores were associated with less severe symptoms of depression (Hawkins et al., 2018)	**1. Self-Compassion Scale Short Form (SCS-SF)** Raes et al (2011)	12 items How I typically act towards myself in difficult times Indicate how often you behave in the state manner 1 (almost never) – 5 (almost always) When I fail at something important to me, I become consumed by feelings of inadequacy I try to be understanding and patient towards those aspects of my personality I don't like When something painful happens, I try to take a balanced view of the situation When I'm feeling down, I tend to feel like most other people are probably happier than I am I try to see my failings as part of the human condition When I'm going through a very hard time, I give myself the caring and tenderness I need When something upsets me, I try to keep my emotions in balance When I fail at something that's important to me, I tend to feel alone in my failure When I'm feeling down, I tend to obsess and fixate on everything that's wrong When I feel inadequate in some way, I try to remind myself that feelings of inadequacy are shared by most people I'm disapproving and judgemental about my own flaws and inadequacies I'm intolerant and impatient towards those aspects of my personality I don't like	12 item measures, good validity and internal consistency in nonclinical samples (Raes et al., 2011) alpha = 0.87 Used in Hawkins et al., 2018 alpha =0.74 Near perfect correlation with long SCS total score r=0.98
Being "battle" ready **Walking the tightrope**	**E. Parent self-efficacy** (Parent beliefs about their capacity to perform in their role as a parent to their child with a visible difference) **Rationale for inclusion:** **Evidence from study 1:** Parents reported being distressed by their perceptions that they did not have	**1. TOPSE Tool to measure Parenting Self-Efficacy** Kendall & Bloomfield (2005) Scale may require adaptation to be relevant to a visible	36 items, 8 subscales Using the scale below, please enter in the boxes how much you agree with each statement. The scale ranges from 0 (completely disagree) – 10 (completely agree) Emotion and affection: I am able to show affection towards my child I can recognise when my child is happy or sad I am confident my child can come to me if they're unhappy When my child is sad, I understand why	Kendall & Bloomfield (2005) Total scale alpha = 0.95 Affection/emotion = 0.81 Play = 0.82 Empathy = 0.89 Routines = 0.84 Control = 0.89 Boundaries = 0.87

	the skills to manage parenting tasks relating to the visible difference e.g., talking to their child about visible difference **Evidence from the literature:** • Low parenting self-efficacy in parents of children and young people with chronic health conditions are associated with psychological distress (Giallo et al., 2013; Harper et al., 2013)	difference population	I have a good relationship with my child I find it hard to cuddle my child Play and enjoyment: I am able to have fun with my child I am able to enjoy each stage of my child's development I am able to have nice days with my child I can plan activities that my child will enjoy Playing with my child comes easily to me I am able to help my child reach their full potential Empathy and understanding**:** I am able to explain things patiently to my child I can get my child to listen to me I am able to comfort my child I am able to listen to my child I am able to put myself in my child's shoes I understand my child's needs Control: As a parent I feel I am in control My child will respond to the boundaries I put in place I can get my child to behave well without a battle I can remain calm when facing difficulties I can't stop my child behaving badly I am able to stay calm when me child is behaving badly Discipline and setting boundaries: Setting limits and boundaries is easy for me I am able to stick to the rules I set for my child I am able to reason with my child I can find ways to avoid conflict I am consistent in the way I used discipline I am able to discipline my child without feeling guilty Pressures: It is difficult to cope with other people's expectation of me as a parent I am not able to assert myself when other people tell me what to do with my child Listening to other people's advice makes it hard for me to decide what to do I can say 'no' to other people if I don't agree with them I can ignore pressure from other people to do things their way I do not feel a need to compare myself to other parents	Pressures = 0.87 Acceptance = 0.93 Learning = 0.81 Test-retest reliability: Affection = 0.75 Play = 0.67 Empathy = 0.58 Routine = 0.74 Control = 0.81 Boundaries = 0.60 Pressures = 0.76 Acceptance = 0.88 Learning = 0.79 Used in the wider paediatric literature: Dai et al (2019) Parents of children with Hirschsprung's disease (a congenital disorder) Zhou et al (2019) Parents of children with autism spectrum disorder

			Self-acceptance: I know I am a good enough parent I manage the pressures of parenting as well as other parents do I am not doing that well as a parent As a parent I can take most things in my stride I can be strong for my child My child feels safe around me Learning and knowledge I am able to recognise developmental changes in my child I can share ideas with other parents I am able to learn and use new ways of dealing with my child I am able to make the changes needed to improve my child's behaviour I can overcome most problems with a bit of advice Knowing that other people have similar difficulties with their children makes it easier for me Possible adaptation of items: **Empathy and understanding:** When talking about my child's condition/injury, I am able to explain things patiently to my child When talking about my child's condition/injury, I can get my child to listen to me I am able to comfort my child when they experience difficulties related to their condition or injury I am able to listen to my child when they talk about their condition or injury When thinking or talking about the condition/injury, I am able to put myself in my child's shoes I understand my child's needs relating to their condition/injury	

Being "battle" ready **Walking the tightrope**	**F. Parent- child communication** (Communication between parent and child when discussing the visible difference: both self-efficacy and style of communication of interest)	**1. Parent-Adolescent communication scale** Barnes & Olsen (1985)	20 items, 2 subscales Please indicate how much you agree or disagree with each of the following statements about general communication between you and your…. Positive (open) I can discuss my beliefs with my *** without feeling restrained or embarrassed. My *** is always a good listener. My *** can tell how I'm feeling without asking	Masselem et al. (1990) Positive > 0.81 Negative > 0.74 Used in wider literature: Parent and adolescents with diabetes

	Rationale for inclusion: Evidence from study 1: Parents are concerned about communicating with their child about the visible difference. They worry that they must get these conversations 'right' in order to inform the child's narrative about their condition. Being confident and feel able to do this may reduce parent distress. **Support from the literature:** • Communication between parents and adolescents for adolescents with Type 1 Diabetes when becoming independent. Parent roles include teaching/educating, reassuring, motivating, conversations about decision making (Tuohy et al., 2019) • SIBS intervention: communication interaction for parents and		I am very satisfied with how my *** and I talk together I openly show affection to my ***. When I asked questions, I get honest answers from my ***. My *** tries to understand my point of view I find it easy to discuss problems with my *** It is very easy for me to express all my true feelings to my ***. Negative (problem) Sometimes I have trouble believing everything my *** tells me I am sometimes afraid to ask my *** for what I want. My *** has a tendency to say things to me which would be better left unsaid. If I were in trouble, I could tell my ***. When we are having a problem, I often give my *** the silent treatment. I am careful about what I say to my ***. When talking to my ***, I have a tendency to say things that would be better left unsaid There are topics I avoid discussing with my ***. My *** nags/bothers me My *** sometimes insults me when she/he is angry with me I don't think I can tell my *** how I really feel about some things.	
		2. Communication in other measures **2.1 Empathy and understanding subscale of TOPSE**	Adapted items: When talking about my child's condition/injury, I am able to explain things patiently to my child When talking about my child's condition/injury, I can get my child to listen to me I am able to comfort my child when they experience difficulties related to their condition or injury I am able to listen to my child when they talk about their condition or injury When thinking or talking about the condition/injury, I am able to put myself in my child's shoes	

	siblings of children with chronic illness, which positive evaluations from children and parents (Vatne et al., 2019) • Improving parent-child communication process may reduce individual health risk factors through discussion about factors that may lead to involvement in health-risk behaviours, such as self-esteem (Reisch et al., 2006)	**2.2 Social situation items from CARe burn scales (See construct C. above)** **2.3 Communication subscale in the parenting inventory measuring stress (See construct B above)**	I understand my child's needs relating to their condition/injury **Not covered by these items:** **How comfortable parents are in passing on information about a child's condition or injury** **Talking about the diagnosis/injury/cause of the visible difference** **Answering questions from the child about the visible difference** **Discussing challenges, the child has encountered**	

Appearance does(n't) matter	**G. Investment in appearance** **Rationale for inclusion: Evidence from study 1:** Professionals reported that parents who appeared to be more investment in their appearance were more distressed by the appearance-affecting aspects of their child's condition e.g., scarring	**1. The Multidimensional Body-Self Relations Questionnaire (MBSRQ)** Cash (2000)	Short form is 34 items 1 (definitely disagree) – 5 (definitely agree) You are asked to indicate the extent to which each statement pertains to you personally. Appearance orientation subscale Before going out in public, I always notice how I look. I am careful to buy clothes that will make me look my best I check my appearance in a mirror whenever I can. Before going out, I usually spend a lot of time getting ready. It is important that I always look good. I use very few grooming products.	Cash (2000) Appearance evaluation Alpha = 0.88 1 month test-rest > 0.81 Used in burn survivors Thombs et al 2008 Connell et al 2013

	Support from theory and literature: • When appearance is more salient, negatively valenced appearance cause greater distress (Moss & Rosser, 2012) • Tripartite model of body image suggests that body appearance ideals from various sources including family can lead to negative outcomes including body image (Thompson et al., 1999)		I am self-conscious if my grooming isn't right. I usually wear whatever is handy without caring how it looks I don't care what people think about my appearance I take special care with my hair grooming. I never think about my appearance I am always trying to improve my physical appearance	

Sample engaged in social support **Walking the tightrope**	**H. Perceived Social Support** (Parent reported social support) **Rationale for inclusion:** **Evidence from study 1:** All parents involved in study 1 were recruited through charity organisations and therefore all engaged with some level	**1. Multidimensional Scale of Perceived Social Support (MSPSS)** (Zimet et al., 1988)	12 items, 3 subscales family, friends and significant other Indicate how you feel about each statement 1 (Very strongly disagree) – 7 (Very Strongly agree) There is a special person who is around when I am in need There is a special person with whom I can share my joys and sorrows. My family really tries to help me. I get the emotional help and support I need from my family. I have a special person who is a real source of comfort to me. My friends really try to help me. I can count on my friends when things go wrong	Zimet et al. (1988) Significant other = 0.91 Family alpha = 0.87 Friends alpha = 0.85 Negatively correlated with anxiety and depression subscales of Hopkins Symptom Checklist (HSCL) Used in wider paediatric literature:

	of social support. Additionally, many reported additional sources of social support. **Support from the literature:** • Parents of children with visible difference report feelings of psychological isolation (Tanner et al., 1998; Heath et al., 2018) • A review of the literature found that parent of children with CLP lack adequate social support (Nelson et al 2012) • Parents of children with congenital appearance-altering conditions benefit from opportunities to access social support (Bogart, 2017) • Approach rather than avoidance orientated coping strategies are associated reduced psychological distress in parent of		I can talk about my problems with my family. I have friends with whom I can share my joys and sorrows. There is a special person in my life who cares about my feelings. My family is willing to help me make decisions I can talk about my problems with my friends.	Grant et al. (2012) Parents of children with an enzyme disorder and children with intellectual disabilities

	children with CLP (Baker et al., 2009)			

Walking the tightrope	**I. Parent treatment knowledge** (Parent self-report knowledge about their child's treatment for their condition or injury) **Rationale for inclusion:** **Evidence from study 1:** Parents reported struggling when they felt they did not have the information they needed to support their child. They also reporting information seeking behaviours and distress when they felt their needs for information or expectations for support were not met. **Support from the literature:** • CEN-Q scores were associated with positive life orientation and greater health-care satisfaction in parents of children with CLP (Stock et al., 2019)	**1. Clinical Excellence Network Questionnaire (CEN-Q)** Stock et al. (2016) Adapt for parent use	5 items Scored 0 (strongly disagree) – 4 (strongly agree) If anyone asks about my condition, I know what to say I have all the information I need about my condition I am happy with the treatment I have received so far I have been involved in decisions about my treatment I would like to have more treatment in the future to improve how I look	Used in visible difference literature: Stock et al (2019) Risk and Protective factors for psychological distress in families of children with CLP

	• Parents of children with CLP report coping by seeking information, emphasising the importance of the availability of accurate information (Camic & Shearer, 2012)			
Appearance does(n't) matter	**J. Perceived severity of difference** (Parent self-report severity of child's visible difference) **Rationale for inclusion: Evidence from study 1:** Parents vary in how noticeable they report their child's visible difference is based on many variables, such as how severe they think it is, how easy it is to conceal, how much they notice it, how much other people comment on it. **Support from the literature:** • Greater perceived/subjective severity is associated with poorer adjustment in individuals with a	**1. 2 items on how noticeable the child's condition/injury is**	**1. How noticeable do you feel your child's condition/injury is?** 0= not at all noticeable…. 10= very noticeable **2. How noticeable do you feel your child's condition/injury is to other people?** 0= not at all noticeable…. 10= very noticeable	

	visible difference (Moss, 2005)			
Appearance does(n't) matter	**K. Appearance fixing behaviours** **Rationale for inclusion: Evidence from study 1:** Parents often reported that they didn't feeling their child's visible difference was noticeable **because** they carried out appearance fixing behaviours e.g., letting their child's hair grow long to conceal Microtia	**1. Body Image Coping Strategies Inventory (BICSI)** (Cash, 2005)	Appearance fixing subscale, 10 items Using the scale below, enter a number from 0 to 3 in the space to indicate how well each way of coping describes what you actually do or would do 0 (definitely not like me) – 3 (definitely like me) I spend extra time trying to fix what I don't like about my looks. I seek reassurance about my looks from other people. I do something to try to look more attractive I spend more time in front of the mirror. I think about what I should do to change my looks. I fantasize about looking different. I think about how I could "cover up" what's troublesome about my looks. I compare my appearance to that of physically attractive people I make a special effort to look my best. I make a special effort to hide or "cover up" what's troublesome about my looks Example adapted items: If I don't like the way my child looks, I spend time trying to fix it I seek reassurance about my child's looks from other people I do something to try to make my child look more attractive	Cash et al (2005) Alphas: Men = 0.91 Women = 0.90 Correlated with measures of salience of appearance and body dissatisfaction Appearance Schemas Inventory – Self-evaluative factor (ASI-R) = 0.32 Body image ideals questionnaire (BIQ) = 0.23 Used in visible difference population: Zuchelli et al. (under review)

Being "battle" ready	**L. Perceived social challenges** (Parent self-report perception of social difficulties being experienced by child)	**1. 3 items on how the parent perception of the current social impact of their child's**	In the past two weeks: **1. How often has your child been teased about the way they look?** 1 (Never) – 5 (Always)	

	Rationale for inclusion: Evidence from study 1: Parents discussed both actual and anticipated social challenges related to their child's visible difference. May be beneficial to know whether the child is currently experiencing social challenges and the parent-reported impact of this on themselves.	**visible difference** Adapted from Project EAT-III Teasing Scale (Neumark-Sztainer et al., 2007) adaptation	**2. How upset were you about your child being teased?** 1 (Not upset) – 5 (very upset) **3. How upset was your child about being teased?** 1 (Not upset) – 5 (very upset)	

Appendix H: Study two materials

Online survey materials

Exploring the experiences of parents and carers of children and young people with an appearance-affecting condition or injury.

Parent and carer information

You are being invited to take part in a research study. Before you decide whether you wish to take part, it is important you know why the research is being done and what it will involve. Please read the information below carefully.

Hello, my name's Maia Thornton and I'm a PhD student at the Centre for Appearance Research (CAR) at the University of the West of England, Bristol.

What is the research about?

Thank you for your interest in this research.

This study will explore the experiences of parents and carers of children and young people with a condition or injury that affects their appearance (e.g., birthmarks, craniofacial and skin conditions, scars from injuries such as burns), and results in what is sometimes called a visible difference or disfigurement. Living with an appearance-affecting condition/injury can present challenges for the child or young person and can also impact other members of the family, including parents and carers.

To find out more about the experiences of parents and carers, I am carrying out a number of research studies. In this online survey you will be asked about the following topics:

- Your experience of parenting a child with an appearance-affecting condition or injury
- How easy or difficult you find interacting with your child in different situations
- How you manage challenges related to your child's condition or injury
- Your thoughts and feelings about your child's condition or injury
- Difficult emotions you might have felt or be feeling

There are no right or wrong answers to the questions. I am interested in hearing your thoughts, so please answer as honestly as you can. If you don’t want to answer a question, for any reason, you can skip it and move on to the next question.

Who can take part?

Any parent or carer of a child or young person (0 - 18 years old) with an appearance-affecting condition or injury can take part. This includes conditions that affect the face or body, skin conditions, conditions or injuries resulting in paralysis of the face or body and changes to appearance as a result of surgery, accident or illness.

Do I have to take part?

No, taking part is entirely voluntary. If you decide that you do not want to take part, this will not be shared with others.

If you change your mind during or after completing the survey and wish to withdraw your data, please contact me at Maia.thornton@uwe.ac.uk and your responses will be deleted. You can withdraw your data for up to two weeks after completing the survey.

Will I be identifiable, will my responses be shown to anyone and who will see them?

Your responses will be anonymous, you will not be asked for your name or any information that will be used to identify you or your child. You will be asked to create a unique ID at the beginning of the study, and this will be used to identify your responses if you wish to withdraw.

Only my Director of Studies (line manager) and I will have access to the original data from the survey. Others will only see the results of the data analysis. We will present demographic data from everyone who takes part in the survey (e.g., sex, age, relationship status, and the nature/location of your child's visible difference) but you will not be individually identifiable.

What are the benefits of taking part?

By sharing your experiences, you will be contributing to the knowledge and understanding of the experiences of parents and carers of children with an appearance-affecting condition or injury. Using the data from this research, we plan to further develop support for parents and families. Your responses will help us to do this.

If you complete the survey, you will be entered into a prize draw to win a £50 Amazon voucher as a thank you for taking part. If you choose to provide an email address to be entered into the prize draw, this information will be stored separately to your survey responses to ensure confidentiality,

Are there any risks involved if I take part?

There are no major risks to taking part in this research. The only thing that we ask you to consider is that the survey may cover topics you find sensitive, such as questions relating to difficult emotions that you might have felt or be feeling and dealing with challenges related to your child's condition or injury.

As a result, there may be questions that you find uncomfortable or upsetting to answer. If you feel unable to answer a question you can skip the question. If you become upset whilst taking part, or do not want to continue for any reason you can stop by exiting the browser page. If you do feel upset after taking part, there will be contact details for relevant support organisations included at the end of the survey. These contact details can also be found the end of this information page.

How will my data be used?

Your data will be used as part of my PhD research. The data will be analysed for my research, and results of the analysis will be reported in my thesis and in any publications and conference presentations arising from my research. The information you provide will only be used for research purposes and to help develop support resources.

If you would like to receive a summary of the findings of the research, there will be an opportunity for you to request this at the end of the survey. If you provide an email address for this purpose, this will be stored separately from your survey responses to ensure confidentiality.

The personal information collected in this research project (e.g., your survey responses) will be processed by the University in accordance with the terms and conditions of the General Data Protection Regulation (2018). We will hold your data securely and not make it available to any third party unless permitted or required to do so by law. If you would like to read the full UWE privacy notice click here.

What if I have questions?

If you have any questions about the research, please contact me at:

Email: Maia.Thornton@uwe.ac.uk

Telephone number: 011732 85817

If you wish to discuss the study with anybody else or if you have complaints connected with the study please contact Dr Heidi Williamson, my supervisor and Senior Research Fellow at CAR. Email: Heidi3.williamson@uwe.ac.uk

Sources of support

If you feel that you need support with any of the topics mentioned, the following websites list some helpful resources:

Mind Mental Health charity

Samaritans charity
Changing Faces charity: supports individuals and families affected by appearance altering conditions and injuries

I am now going to ask you to provide consent to take part in the study.

Please read the statements and respond **yes if you agree** and **no if you do not.**

	Yes	No
I have read the information page about the study.		
I understand the information sheet and I have had the opportunity to contact the research team and ask questions about it.		
I understand that taking part will involve answering questions about my experience of caring for a child with an appearance-affecting condition or injury.		
I understand that my participation is voluntary, and I can stop the survey at any point if I no longer wish to take part.		
I understand that I can skip any questions that I do not want to answer.		
I agree to the University of the West England processing my data as described in the privacy notice within the information sheet.		
I understand that I can withdraw without providing a reason, and with no consequences, for up to two weeks after completing the survey (by emailing the researcher).		
I consent to participating in this research.		

Thank you for agreeing to take part in this study.

Firstly, I would like to ask you some questions to confirm whether you are eligible to take part in the study.

	Yes	No
Does your child (a child for which you have caring/guardianship responsibilities) have a condition which affects the way they look?		
Is that child 18 or under?		
Do you live in the UK?		
Do you have a good understanding of written English?		

Thank you for answering these questions, you are eligible to take part in the survey.

This survey will ask about:

- Your child's condition or injury, with a particular focus on the aspects that affect their appearance

- Different situations related to your role as a parent of a child with an appearance-affecting condition or injury
- How you respond to potentially difficult situations as a parent

The survey should take between **20 and 30 minutes** to complete.

If you complete the survey, you will have the opportunity to be entered into a prize draw to win a **£50 Amazon e-voucher**, as a thank you for your time.

You will also have the opportunity to request a summary of the results of this study.

Please remember that there are no right or wrong answers, you are the experts of your own experience. Take your time and please answer as honestly as possible.

The first thing you will need to do is create a **unique participant code.**

As your answers are anonymous this code will be used to identify your response if you wish to withdraw your data.

First, please enter the number of the day you were born e.g., if you were born on the 11th of January, you would enter 11.

Now, please enter the last letter of your first name e.g., if your name was Alice you would enter E.

Finally, please enter the first letter of the name of the place you were born e.g., if you were born in London you would enter L.

Thank you for creating your unique participant code.

I would now like to ask you a few questions about yourself. Your answers will help me to understand a bit about your background and your family.

How old are you?

What is your gender?

Female	
Male	
Non-binary	
Prefer to self-describe	
Prefer not to say	

How would you describe your ethnic background?

White
English/Welsh/Scottish/Northern Irish/British
Irish
Gypsy or Irish Traveller
Any other White background

Mixed/Multiple ethnic groups
White and Black Caribbean
White and Black African
White and Asian
Any other Mixed/Multiple ethnic background

Asian/Asian British
Indian
Pakistani
Bangladeshi
Chinese
Any other Asian background

Black/African/Caribbean/Black British
African
Caribbean
Any other Black/African/Caribbean background

Other ethnic group
Arab
Any other ethnic group

What is the highest level of educational or school qualifications that you have completed?

No qualifications	
Level 1 qualifications/ Level 1 BTEC/ Level 1 NVQ or equivalent	

Level 2 qualifications/ GCSEs / Level 2 BTEC/ Level 2 NVQ or equivalent	
Level 3 qualifications/ A levels/ Level 3 BTEC/ Level 3 NVQ or equivalent	
First degree e.g. Bachelor's degree or equivalent	
Master's degree	
Doctorate degree	

How would you describe your marital status?

Single	
Married/Civil Partnership	
Divorced	
Separated	
Cohabitating	
Other	

How many children do you have?

What are the ages of your children?

What is your relationship to the child with the appearance-affecting condition or injury?

I would like to ask you a few more questions about your child, so that I can better understand your experience as a parent or carer.

For the rest of the questions, I would like you to answer thinking about one child in your family who has an appearance-affecting condition or injury.

What condition or injury does your child have?

Please describe how the condition or injury affects their appearance.

How old is your child?

What is their gender?

Female	
Male	
Non-binary	
Prefer to self-describe	
Prefer not to say	

Do you have an appearance-affecting condition or injury?

If yes, what condition or injury do you have? And how does it affect your appearance?

The next couple of questions will ask you to think about how easy it is to notice your child's condition or injury.

On the scale below please indicate how **noticeable** your child's condition or injury is **to you**

Not at all noticeable Very noticeable

0 10

On the scale below please indicate how **noticeable** your child's condition or injury is **to other people**?

Not at all noticeable Very noticeable

0 10

In the next few questions please tell me how you feel about any treatment your child has received and any information you have had about their condition or injury.

	Strongly Disagree 0	Disagree 1	Neither agree nor disagree 2	Agree 3	Strongly agree 4	Not Applicable
If anyone asks about my child's condition, I know what to say						
I have all the information I need about my child's condition						
I am happy with the treatment my child has received so far						
I have been involved in the decisions about my child's treatment so far						
I would like my child to have more treatment in the future to improve how they look						

The next series of questions will be about how you communicate with your child and others about their appearance-affecting condition or injury.

	Never 1	Sometimes 2	Often 3	Most of the time 4	Always 5
I feel ok answering my child's questions about their difference in appearance					
I feel ok talking to my child about the reason they look different					
I feel ok talking to my child about difficulties relating to their appearance difference					
I feel ok talking to my child about treatment that will alter their appearance					

Firstly, I would like you to think about how you communicate with <u>your child about their condition and injury.</u>

Please read the statements below and answer thinking about how you have felt over the last week.

Next, I would like you to think about how you communicate about your child's condition or injury to other people.

Please read the statements below and answer thinking about how you have felt over the last week.

	Never 1	Sometimes 2	Often 3	Most of the time 4	Always 5
I feel ok when **other people** look at my child's condition or injury					
I feel ok when **other people** ask about my child's condition or injury					
I feel ok talking to **other people** about my child's condition or injury					

Do you have anything else you would like to share relating to communication about your child's condition or injury?

For the next series of questions, please think about how people respond to your child's difference in appearance.

Firstly, I would like to hear about the way you respond to your child's appearance difference

Parents and carers have different responses to their child looking different. It is interesting for me to hear about this so that I can better understand your experience.

Please read each statement and think about whether it is characteristic of how you might respond to your child's difference in appearance.

	Definitely not like me 0	Mostly not like me 1	Mostly like me 2	Definitely like me 3
I cover up or hide my child's difference in appearance.				
I spend time thinking about how to alter my child's appearance.				
I avoid situations where my child's difference in appearance might be seen.				

I try not to talk or think about my child's difference in appearance.				

Next, I would like to hear about how you think other people respond to your child's difference in appearance.

Please read each question and answer thinking about the last two weeks

	Never 1	2	Sometimes 3	4	Always 5
How often has your child been teased about the way they look?					
	Not upset 1	2	Somewhat upset 3	4	Very upset 5
How upset were **you** about your child being teased?					
How upset was **your** child about being teased?					

Do you have anything else you would like to share related to your own or other people's responses to your child's difference in appearance?

For the next series of questions, I will be asking you to think about how you feel as a parent.

Please read the words below and think about how much you have felt this way in your role as a parent of a child with an appearance-affecting condition or injury.

Please think about the way you have felt over the last week.

	Very slightly of not at all	A little	Moderately	Quite a bit	Extremely
Interested					
Distressed					
Excited					
Upset					
Strong					
Guilty					

Scared					
Hostile					
Enthusiastic					
Proud					
Irritable					
Alert					
Ashamed					
Inspired					
Nervous					
Determined					
Attentive					
Jittery					
Active					
Afraid					

Now could you please read the following statements and think about <u>how frequently</u> you worry about these situations.

	Never 1	2	Sometimes 3	4	Very often 5
Worrying about the long-term consequences of the condition or injury					
Feeling helpless regarding my child's situation					
Feeling uncertainty about the future					

I'd like to ask you about the same situations

as above, but this time please read each statement and think about <u>how difficult</u> you find each situation to deal with.

	Not at all 1	2	Sometimes 3	4	Extremely 5
Worrying about the long-term consequences of the condition or injury					
Feeling helpless regarding my child's situation					
Feeling uncertainty about the future					

Do you have anything else you would like to share related to <u>your feelings as a parent?</u>

You're half way through! Thank you so much for answering the questions so far. When you are ready continue on to the next section.

For the next series of questions, I will be asking you to think about the social support that you receive if/when you experience challenging situations in <u>your role as a parent of a child with an appearance-affecting condition or injury.</u>

Please read the statements below and indicate how you feel about each statement.

	Very Strongly Disagree 1	Strongly Disagree 2	Mildly Disagree 3	Neutral 4	Mildly Agree 5	Strongly Agree 6	Very Strongly Agree 7
There is a special person who is around when I am in need							
There is a special person with whom I can share my joys and sorrows							
My family really tries to help me							
I get the emotional help and support I need from my family							
I have a special person who is a real source of comfort to me							
My friends really try to help me							
I can count on my friends when things go wrong							
I can talk about my problems with my family							
I have friends with whom I can share my joys and sorrows							
There is a special person in my life who cares about my feelings							
My family is willing to help me make decisions							
I can talk about my problems with my friends							

Do you have anything else you would like to share related to <u>social support</u> you receive?

Thank you so much for helping me with my research, not many questions left now.

For the next series of questions, can you tell me <u>how you typically act towards yourself in difficult times?</u>

Please read each of the statements below and respond by selecting how often you would typically respond in these ways.

	Almost Never 1				Almost Always 5

When I fail at something important to me, I become consumed by feelings of inadequacy					
I try to be understanding and patient towards those aspects of my personality I don’t like					
When something painful happens, I try to take a balanced view of the situation					
When I’m feeling down, I tend to feel like most other people are probably happier than I am					
I try to see my failings as part of the human condition					
When I’m going through a very hard time, I give myself the caring and tenderness I need					
When something upsets me, I try to keep my emotions in balance					
When I fail at something that’s important to me, I tend to feel alone in my failure					
When I’m feeling down, I tend to obsess and fixate on everything that’s wrong					
When I feel inadequate in some way, I try to remind myself that feelings of inadequacy are shared by most people					
I’m disapproving and judgemental about my own flaws and inadequacies					
I’m intolerant and impatient towards those aspects of my personality I don’t like					

As above, please read each of the statements below and respond by selecting how much they reflect the way you feel.

	Strongly disagree	Disagree	Neutral	Agree	Strongly agree
In uncertain time, I usually expect the best					
It’s easy for me to relax.					
If something can go wrong for me, it will.					
I’m always optimistic about my future.					
I enjoy my friends a lot.					
It’s important for me to keep busy.					
I hardly ever expect things to go my way.					
I rarely count on good things happening to me.					
Overall, I expect more good things to happen to me than bad.					
I don’t get upset too easily.					

Do you have anything else you would like to share related to coping with difficult situations?

The next set of questions will ask you to think about you think and feel about different situations in your role as a parent of a child with an appearance-affecting condition or injury.

Please read each of the statement and respond by rating how true you feel each statement is for you.

	Never true	Very seldom true	Seldom true	Sometimes true	Frequently true	Almost always true	Always true
My emotions get in the way of being the type of parent I would ideally like to be.							
My worries get in the way of me being successful as a parent.							
My emotions cause problems in my relationship with my child.							
It seems to me that most people are better parents than I am.							
My painful memories prevent me from parenting the way that I would like.							
My feelings stop me from doing what I know is best for my children.							
I worry about not being able to control the feelings I have about my children.							
I have to feel in the mood before I can give my child affection or attention.							
I could not cope with the guilt if my child did something wrong.							
I don’t let my child do many things with their friends because I don’t think I could cope if something bad happened to him/her.							

I have refused to let me child do things that were important to them because I would worry too much (e.g., spend time with friends, walk to school by themselves).							
I don’t let my child do things that I’ll worry about.							
I am responsible for my child’s behaviour.							
I can still take care of my parenting responsibilities even when I feel tired, stressed, sad or angry.							
I can get angry with my children and still be a good parent.							
I can have a good relationship with my children no matter what I am thinking and feeling.							
Watching my child deal with new experiences as he/she grows up (e.g., starting high school, first kiss, puberty) is interesting and exciting.							
I am able to separate how I respond to my children from how I am feeling.							
The unpredictability of being a parent is one of the things that makes parenting fun and rewarding.							

For the next series of questions, I will be asking you to think about how confident you feel in different situations in your role as a parent or carer of a child with an appearance-affecting condition or injury.

	Not at all confident							Very confident		
	1	2	3	4	5	6	7	8	9	10
I can support my child in telling others about their condition or injury.										
I can support my child to socialise with other children their age.										
I can find the information I need to support my child.										
I can find additional support for my child's wellbeing, if they need it.										
I can make decisions about treatment for my child's condition or injury.										
I can support my child in making decisions about treatment for their condition or injury.										
I can help my child with their treatment for their condition or injury.										
I tend to know if my child is upset about their appearance difference.										
I can help my child to feel comfortable about their appearance.										
I can promote confidence and resilience in my child.										
I can help to prepare my child for difficulties they may come across in life.										

Do you have anything else you would like to share related to how confident you feel in your role as a parent of a child with an appearance-affecting condition or injury?

For the next series of questions, I will be asking you to think about your own appearance.

Please read the statements below and indicate the extent that each statement is relevant to you.

	Definitely disagree	Mostly disagree	Neither agree nor disagree	Mostly agree	Definitely agree

	1	2	3	4	5
Before going out in public, I always notice how I look					
I am careful to buy clothes that will make me look my best					
I check my appearance in a mirror whenever I can					
Before going out, I usually spend a lot of time getting ready					
It is important that I always look good					
I use very few grooming products					
I am self-conscious if my grooming isn't right					
I usually wear whatever is handy without caring how it looks					
I don't care what people think about my appearance					
I take special care with my hair grooming					
I never think about my appearance					
I am always trying to improve my physical appearance					

Do you have anything else you would like to share about your thoughts or feelings about your own appearance?

Last question, is there anything you would like to add about any of the topics covered in this survey?

Thank you for taking part!

I really appreciate you giving your time to support this research. Your responses will help me to better understand the experience of caring for a child with an appearance-affecting condition or injury. This will help me to develop future support for parents and carers.

If you would like to be entered into the prize draw to win a £50 Amazon voucher, please enter your email address below

Would you like to receive a summary of the results of this study?

Yes	
No	

If you have ticked yes, please provide an email address below that you would like the summary to be sent to.

Sources of support

If any of the topics covered in the survey have left you feeling that you might need some additional support, please take a look at the resources listed below:

Mind Mental Health charity: https://www.mind.org.uk/

Samaritans charity https://www.samaritans.org/

Changing Faces charity: supports individuals and families affected by appearance altering conditions and injuries:

Website: https://www.changingfaces.org.uk/

Advice and support information line: 0300 012 0275 (open Mon-Fri 10am-4pm)

Data cleaning and screening plan

Data task	How?
Initial screening of responses	• Check for any response with durations that are largely different to expected values • Check for participants from outside the UK • Check for participants who display suspicious response patterns • Check details about child appearance difference to ensure they have a visible difference • Check parent or carer role/status Remove any suspicious responses from data set e.g., attention checks, very fast response times, duplicate responses
Separate qualitative data from quantitative data	• Remove qualitative data from open ended questions from main data set • Save to a separate data file for content analysis at a later time
Code the data set in preparation for analysis	• Follow codebook in order to code data correctly and prepare for analysis
Check for errors	• Check for scores that fall out of the possible range and correct any errors (use descriptives on SPSS) - Check minimum and maximum values on each variable to ensure they are within possible range - Check the mean scores on each variable, does it make sense? - Check for duplicate or missing cases
Check for missing data and missing data patterns	• There are very few small amounts of missing data – mostly due to parents not feeling that questions are relevant to them and their children (e.g., very young children) • Need to determine what % of individual variables is missing (if under 5% it's fine – I suspect that it is) - Run frequencies to identify % of missing values - Conduct a pattern analysis to confirm whether any missing data on different variables is related - Code missing values in the variable view and insert coded values into data view

Checking the normality of distribution of scores	• Important for many statistical techniques so worth doing in these initial stages • Explore option on descriptives (select descriptives and outliers) • Normality plots (select Histogram & normality plots with tests) • This will later be followed by checking specific assumptions for multiple regression

Content analysis exemplar quotes

Theme	Sub-theme	Exemplar quote
Reactions to visible difference	Emotional impact on parents	“I feel guilty that people see his inflamed skin and assume I'm doing something wrong, mostly because I feel guilty that I could be doing more for him”
	Lack of public awareness	“It is usually out of ignorance that people make comments. It’s frustrating that there is not better education and subsequently acceptance of facial differences.” “We often educate both healthcare professionals and other non healthcare workers about it when asked” “I would prefer adults to ask rather than point and stare”
	Impact on children	“My child even at only 4 understands that a lot of people point and laugh/make nasty comments. And gets very upset about it and even refuses to wear clothes that reveals to much of his marks.” “He [sibling] can sometimes adopt the role of parent and take on too much.”
	Visibility	“My son gets looked at alot because of his birthmark being so visible on his face.”
	Too young	“I'm not sure he's old enough to be teased quite yet.”
Communication	Acceptance	“We have brought her up to embrace her difference, however, with her only being 4 this is an ongoing process.”
	Preparing their child	“We teach her everything we know about vitiligo, so that she too can educate those around her as honestly as possible” “I let my son figure it out for himself. It’s hard as I want to jump in every time he struggles but he has great determination and doesn’t give up” “I think it is important to be open and upfront about how her condition has affected her.”
	Too young	“As he is not yet 3 years old and the birthmark is on his shoulder blade I'm not sure how aware he is of it.”
Affect/stress	Affect	“It took until he was about 10 years old for me to stop wishing it away every single day.”

		“I feel proud taking about the surgery he went through and how he coped.”
	Guilt	“I cannot help but feel guilty at the same time as it makes me think of the accident and our time in hospital.”
	Anxiety	“As her parent I want to protect her from all negativity that she may face, and it makes me a little anxious to think other children and their parents may be judging her appearance”
		“The unknown is always scary and with any diagnosis there is at least some aspects that are unknown as each person is different”
	Stressful situations	“Exhausting - we have to stick strictly to a daily routine of creams, baths, bandages etc for my daughter and this can be tiring.”
Coping	Active coping	“I have spent a lot of time researching my daughter’s condition.”
		“When I am anxious, it helps to get outdoors for a long walk.”
		“I set up a national support group and charity so people would have the support they needed when their babies were born.”
	Psychological resource	“I am really working on trying to be more compassionate with myself and my own failures and not catastrophize as much in uncertain/bad times.”
		“I am naturally a very positive and enthusiastic person.”
	Social support	“With time, family and friend support and help from the charity I feel much more able to cope and help my daughter now.”
		“I appreciate attending conferences etc when you hear from other families and professionals supporting families in their jobs to learn more.”
Self-efficacy	Confidence in skills and knowledge	“But I am prepared to answer his questions and talk to him and other freely about difficulties he may have or potentially have.”
	Weight of responsibility	“I often think as she gets older, I need to be ready to support her for when this happens”
	How do I do this?	“I don’t know how to advise her to respond to cruel remarks well enough”

		“I feel helpless as I do not know what can be done to change her situation as she expresses that she wishes to have it removed but I do not know if it is possible to do so and what the consequences would be.”
Appearance investment	Not important to me	“It is not something I think about a lot.”
	Appearance is important	“Despite trying to change my feelings, I definitely still feel that thin, blonde, pretty is best.”
	Modelling positive body image to child	“I want to build a more confident self-image because I want my daughter to see that and be confident in herself as well.”
Impact of COVID	Support	“COVID-19 has really limited the amount of practical help / face to face support that I have been able to ask for.”
	Social contact	“I worried during lockdown that not socialising with her friends and going to nursery would set her back in this regard.”
		“Because of lockdown, he hasn’t had any contact with other children in last two weeks.”
		“We have been following the pandemic social distancing protocols so have not been in any situations where he could have been teased.”
	Positive experiences	“Lovely to have the children at lunch and supper each day as a family”
	Health concerns	“The COVID-19 pandemic has truly thrown our plans into a tailspin. I’m concerned for her health and my entire family’s health - especially if something were to happen to me or my husband and providing for her once we have passed away”

Appendix I: Mixed methods joint display

Mixed methods joint display

Qualitative themes/subthemes Study 1	**Summary of study 2 findings**	**Level of agreement**	**Mixed methods interpretation**
Theme 1: Appearance does(n't) matter **Subtheme 1: "it broke me"**			
- **Parents talked about the distress they experienced related to their child's visible difference.** - **Parents talked about initial feelings of shock, anxiety and low mood in response to seeing the impact of the condition/injury on their child's appearance** - **For some parents this was a short-term reaction to the visible difference** - **For others it persisted for years** - **Challenged hopes and dreams of the 'perfect' child** - **Feelings of loss and guilt**	Negative relationship between child age and negative affect $p <$ **0.05** The findings suggest that some parents can experience negative emotions, particularly when the child is younger (during the first few years of life). Negative relationship between child age and positive affect $p <$ **0.01** Relationship between child age and positive affect partially mediated by teasing. However, parents do report concerns that as their children grow up and become more independent, they may be	Partial agreement The findings agree that some parents experience distress associated with their child's visible difference in the first few years of life. However, parents may also experience distress as their child grows up and is exposed to increased social challenges e.g., teasing.	Some parents do experience psychological distress related to their child's visible difference. [QUAL + QUANT] As the child grows up, different challenges may trigger difficult and challenging emotions in parents [QUANT]. Parents reported experiencing anxiety, low mod, guilt and loss [QUAL].

	exposed to greater social challenges. Positive and negative affect exist on separate continuums. So, it is possible that some things get easier as a child gets older, whereas others things become more difficult. Parents may experience both positive and negative affect simultaneously.		
Theme 1: Appearance does(n't) matter **Subtheme 2: "They become so much more"** • **For some initial feelings of shock or distress faded over time** • **As their child grew up/time moved on, appearance became a less important aspect of the child's identity** • **Some parents felt there was nothing wrong with their child and no need to alter anything about them** • **After initially experiencing difficult feelings, some parent rebuilt and adjusted**	Negative relationship between child age and negative affect **$p < 0.05$** Negative relationship between child age and positive affect **$p < 0.01$** Negative relationship between appearance investment and negative affect **$p < 0.05$** There were differences in how parents understood appearance investment. Some viewed investment in appearance more	Partial agreement In some aspects, both sets of findings indicate that distress associated with the child's visible difference may lessen as they grow up. However, other findings suggest that distress can persist or increase as they child gets older. Positive and negative affect exist on separate continuums. So, it is possible that some things get easier as a child gets older, whereas others things become	Some parents do experience psychological distress related to their child's visible difference. [QUAL & QUANT} As the child grows up, different challenges may trigger difficult and challenging emotions in parents [QUANT]. As their child grows up other aspects of their child's identity become more important than their appearance difference [QUAL].

• **Parents varied in their judgements about the importance of appearance** • **Broader positive outcomes – greater acceptance of appearance diversity, empathy for others and appreciation of resilience**	in terms of investment in their physical and mental health. Other parents spoke about using appearance fixing behaviours to cope with difficult feelings. This suggests a bidirectional relationship. Negative relationship between optimism and negative affect $p < 0.1$ Negative relationship between optimism and stress frequency $p < 0.2$ Some parents had a more optimistic outlook about their child's future than others. This may contribute to them experiencing less negative affect and stress.	more difficult. Parents may experience both positive and negative affect simultaneously. A possible bi-directional effect indicates a possible relationship between appearance investment and negative affect. But the causal direction is unclear.	Parents reported broader positive outcomes associated with caring for a child with a visible difference [QUAL]. Some parents were more optimistic than others about their ability to adjust and adapt and an optimistic outlook appearance to be protective against distress and stress [QUAL & QUANT].
Theme 2: Being "battle" ready **Subtheme 1: Identify the threats** • **Parents worried most about the social impact of the child's visible difference** • **Concerns about social stigma, discrimination**	Positive relationship between teasing and stress frequency **$p < 0.01$**	Partial agreement Although both data sets found that social challenges were distressing to parents, gender of	Actual and anticipated social challenges related to their child's visible difference may result in distress and stress for parents [QUAL & QUAL].

and the possible negative impact that this might have on their child • **Parents discussed gender differences in social impact, with appearance more salient for girls and societal expectations of appearance more closely tied to worth**	Positive relationship between teasing and stress difficulty **$p < 0.01$** Some parents added that their child was too young to experience teasing. Some of these parents spoke about being worried about their child being teased or bullied as they grew up. Parents reported their child’s reduced social interactions with peers and members of the public, as a result of COVID-19.	child was not a significant predict of distress or stress in parents Silence on QUAL	Parents talked about social challenges potentially being more impactful for girls [QUAL].
Theme 2: Being “battle” ready **Subtheme 2: Shielding my child** • **Attempts to shield their child from potential threats** • **Altering appearance** • **Stepping in or sheltering child from social interactions** • **Parents may choose to conceal their child’s visible difference to manage their own anxiety**	Positive relationship between noticeability to parent and stress frequency **$p < 0.01$** Positive relationship between appearance fixing behaviours and stress difficulty **$p < 0.05$** The qualitative data from the survey found that parent appearance fixing behaviours do seem to be motivated by fear and anxiety about their child’s visible difference being visible and what	Agreement	Parents of children with visible difference may use appearance fixing behaviours to negate their concerns and stress relating to their child’s visible difference and the possible impact of social challenges. [QUANT & QUAL]

	the consequences of this would be. Some parents reported that social isolation as a result of COVID-19 had the benefit of shielding their child from unwanted attention.	Silence on QUAL	
Theme 2: Being "battle" ready Subtheme 3: Arming my child • **Parents talked about the need to arm their child with psychosocial tools to self-manage challenges** • **Building confidence and resilience** • **Weight of responsibility – had they done enough** • **Communicating with child about visible difference and diversity more broadly** • **Importance of modelling adaptive responses to child**	Negative relationship between parent-child communication and negative affect $p < 0.05$ Negative relationship between parent-child communication and stress difficulty $p < 0.05$ Parent report efforts to be open and communicative with their children and this seems to have a positive impact on the parent's feelings of the visible difference – feeling happier and more confident that their child is equipped. Positive relationship between social confidence and positive affect $p < 0.05$	Agreement	Parents are aware of the need to prepare their child to manage situations related to their visible difference independently [QUAL & QUANT] There is a significant negative relationship between effective parent-child communication regarding their visible difference and negative affect [QUANT]. Similarly, there is a relationship between social confidence in parents and positive affect [QUANT]. Parents a desire to model adaptive responses to situations related to their child's visible difference [QUAL].

	Parents also focus on modelling behaviours to their children and providing them with information to handle conversations with others about their visible difference.		
Theme 3: Walking the tightrope Subtheme 1: "Messing about in the dark" - **Parents feeling lost about the best way to support their child** - **Striking the balance with appearance talk** - **A lack of clarity and confidence in parent also observed by professionals**	Positive relationship between self-efficacy and positive affect $p < 0.01$ Some felt positive and assured whereas others lacked confidence and wanted more support. Parents who felt less confident also reported feeling anxious or helpless. Negative relationship between parent-child communication and negative affect $p < 0.05$ Negative relationship between parent-child communication and stress difficulty $p < 0.05$	Agreement	Some parents struggle with a lack of confidence around how to best support their child [QUAL & QUANT]. Significant positive relationship between parent-reported self-efficacy and positive affect suggests that higher self-efficacy promotes positive affect [QUANT]. One key aspect of this confidence around the balance of appearance talk with their child [QUAL]. The significant negative relationship between effective parent-child communication and stress difficulty and negative affect suggests that effective communication may be

			protective against parent distress and suggest [QUANT].
Theme 3: Walking the tightrope Subtheme 2: The double-edged sword of support • **Mixed experiences when reaching out for support** • **Parents can feel disappointed or dismissed after interactions with health professionals** • **Professionals talked about a lack of confidence when working with those with appearance-affecting conditions** • **Parents sought information and support online, again with mixed results** • **Peer support platforms were useful to share experiences, reduce feelings of isolation** • **Parents and professionals also spoke about how peer support can fuel anxiety and expose parents to worst case scenarios**	Negative relationship between knowledge and satisfaction and negative affect **$p < 0.05$** Negative relationship between knowledge and satisfaction and stress frequency **$p < 0.01$** Parents placed importance on being well informed about their child's condition/injury. Positive relationship between social support and stress frequency $p < 0.2$ Positive relationship between social support and stress difficulty $p < 0.2$ Parents value social support but there seem to be many cases where social support is not meeting the needs of the family.	Agreement Silence on QUAL	It is important to parental wellbeing that they feel informed about their child's condition/injury [QUAL & QUANT] A significant negative relationship between knowledge and satisfaction and negative affect and stress suggests that high knowledge and satisfaction may be protective against parental distress and stress [QUANT]. Parents reported mixed experiences of social support. The presence of social support is not sufficient to meet the needs of parents, the quality of the social support is equally important [QUAL & QUANT]. Parents were not always satisfied with their interactions with healthcare professionals, and this

	Access to healthcare and social support affected by COVID-19.	Silence on QUAL	sometimes led to anxiety or stress [QUAL]. COVID has had a negative impact on parent satisfaction with their child's care [STUDY 2 QUAL].
Theme 3: Walking the tightrope Subtheme 3: "On the edge of a big black hole" • **Parents felt fully responsible for their child's ability to cope with challenging situations and carried guilt if their child struggled to adjust** • **Detrimental impact on psychological wellbeing of the parent**	Negative relationship between self-compassion and stress frequency ***p* < 0.05** Negative relationship between self-compassion and stress difficulty ***p* < 0.01** Self-compassion mediates the relationship between psychological flexibility and stress difficulty Parents who are able to take a self-compassionate approach may experience less distress and stress. Negative relationship between optimism and negative affect $p < 0.1$	Agreement	Parents felt guilt and anxiety if their child struggled to adjust or if they had concerns about what would happen in the future [QUAL & QUANT]. Parents were also worried about their own ability to cope if their child struggled [QUAL]. Parent who were able to sit with these difficult feelings and practice self-compassion when things did go wrong, were less likely to experience stress [QUAL & QUANT]. Some parents were more optimistic than others about their child's ability to adjust and adapt and an optimistic outlook appearance to be protective

	Negative relationship between optimism and stress frequency $p < 0.2$ Some parents had a more optimistic outlook about their child's future than others. This may contribute to them experiencing less negative affect and stress.		against distress and stress [QUAL & QUANT].

Appendix J: Intervention content breakdown

Intervention section	Content	Theoretical concepts
Introduction	**Aims:** To introduce the materials and the briefly describe the sections and content. To familiarise parents with the concept of parent-child modelling. **Content breakdown:** • Background on who the intervention materials are for • A summary of research findings of parents of children with visible differences • A summary of the main areas covered • Introduction to parent-child modelling • Tree metaphor (building a strong foundation for child to go on and manage independently) • The structure of the materials	Parent-child modelling
Difficult thoughts and feelings	**Aims:** Introduce the concepts of thoughts and feelings and why we experience them Introduce cognitive defusion, experiential avoidance and value-based action **Content breakdown:** • What are thoughts and feelings? • A summary of parent research findings about thoughts and feelings related to their child's visible difference • An evolutionary explanation for thoughts and feelings – highlight self-compassion when experiencing difficult thoughts and feelings • Introduction to cognitive fusion and defusion ("getting hooked" and "unhooking") • Defusion exercise	Cognitive defusion Experiential avoidance Values-based action Self-compassion

	• Introduction to experiential avoidance – beach ball metaphor • Impact of thoughts and feelings on our behaviour – passengers on the bus metaphor	
Helper skills	**Aims:** Introduce concept of helper skills: being present and knowing your values Provide examples of useful exercises to practice helper skills **Content breakdown:** **Being present:** • Introduction to being present and why this important • Cognitive defusion techniques (I'm having the thought that…, write it down) • Mindfulness techniques (breathing exercises, dropping anchor technique) **Knowing your values:** • Introduction to values and why they are important • Value clarification exercise • Value-based action • Committed action exercise with self-compassion	Cognitive defusion Mindfulness Committed action Self-compassion
Parent-child communication	**Aims:** Explore parent-child communication in-depth Provide parents with guidance and practical skills for communicating with their child and others about their visible difference **Content breakdown:** • Summary of the literature related to parent-child communication in the visible difference field • Discuss parent-child modelling in terms of communication • Practical advice about having conversations about visible difference • Storytelling exercise	Parent-child communication Parent-child modelling Revisiting helper skills Self-compassion

Other people's reactions	**Aims:** Explore the experience and impact of other people's reactions to the child's visible difference. Provide parents with guidance and practical skills for managing the impact of other people's reactions. Provide guidance on how inform other important adults in your child's life about their condition or injury **Content breakdown:** • Managing other people's reactions (Explain, Reassure, Distract, managing staring) • Teasing and bullying (advice for child and parent and self-compassion for parent) • Communicating with other people about the condition or injury (e.g., teachers) • Communication template • Revisiting helper skills and self-compassion for difficult thoughts and feelings	Social challenges Revisiting helper skills Self-compassion
Staying informed	**Aims:** Explore information seeking methods. Provide advice and guidance for potentially anxiety inducing information seeking situations e.g., hospital appointments. **Content breakdown:** • Introduction to why it can feel important to feel informed about child's condition/injury • Practical advice about attending appointments – revisiting helper skills • Normalising difficult thoughts and feelings in challenging situations and not being able to know everything – self-compassion • Appointment Q and A template	Revisiting helper skills Self-compassion
Social support	**Aims:** Explore the benefits of finding the social support that meets their needs Provide parents with guidance and advice on how to set boundaries around support and where to find support online.	Social support Revisiting helper skills Self-compassion

	Content breakdown: • Introducing the benefits of effective social support • Research summary of social support in visible difference • How to identify own social support needs • Setting boundaries around support – include self-compassion and helper skills • Online sources of support	

Appendix K: Participatory Action Research

Demographic information for public involvement workshops

Table 20: Demographic information of parent user representatives

	Mean	SD
Age of parent	49.25	13.55
Age of child	11.40	6.09
	N	**%**
Gender		
Female	5	100
Male	0	0
Child Gender		
Female	2	40
Male	3	60
Relationship to child		
Mother	5	100
Father	0	0
Ethnicity		
White British	4	80
Did not report	1	20
Level of Education		
GCSEs	2	40
A Levels/HND/BTEC	0	0
Bachelor’s degree	1	20
Master’s degree	0	0
Doctorate degree/PhD	0	0
Did not report	1	20

Table 21: Demographic information of health and support professionals

	Mean	SD
Age	38.67	10.26
	N	**%**
Gender		
Female	3	100
Male	0	0
Ethnicity		
Asian/Asian British	1	33
White British	2	66
Level of Education		
GCSEs	0	0
A Levels/HND/BTEC	0	0
Bachelor’s degree	1	33
Master’s degree	1	33
Doctorate degree/PhD	1	33
Job Role		
Charity managerial and support	1	33
Clinical Psychologist	1	33
Audiologist	1	33

Public involvement workshop discussion guide

Can you tell me about your overall impressions of the parenting toolkit?

How did you find the online workbook format?

Introduction:

- Did you feel the introduction explained the structure of the materials?
- Did you feel the introduction explained the rationale for the content included?
- Did you feel you understood who the materials were for?
- Was there anything that was difficult to understand of confusing?

Section one:

Difficult thoughts and feelings:

- Do you feel that the concepts of thoughts and feelings were clearly explained?
- Do you feeling the concept of "getting hooked" and "unhooking" was clearly explained?
- Did you feel that the metaphors used in this section (e.g., passengers on the bus) were clear?
- Do you feel that the examples used were relevant?

Helper skills:

- Did you feel that you understood what helper skills were?

Being present:

- Did you feel that "I am having the thought that…" technique was easy to understand?
- Did you try this technique yourself? If so, how did you find it?
- Did you feel that the breathing technique was easy to understand?
- Did you try this technique yourself? If so, how did you find it?
- Did you feel that the dropping anchor technique was easy to understand?
- Did you try this technique yourself? If so, how did you find it?

Knowing your values:

- Did you feel that the concept of values was explained clearly?
- Did you feel that the family values exercise was easy to understand?
- Did you try this technique yourself? If so, how did you find it?
- Did you feel that the concept of value-based action was explained clearly?
- Did you feel that the value-based action exercise was easy to understand?
- Did you try this technique yourself? If so, how did you find it?

Summary:

- Do you feel the section summary includes everything that it should?
- Would you add anything to the summary?

Section two:

- Do you feel this section covers the main topics that are relevant to parents?
- Is there anything missing that you feel is important?

Talking about your child’s condition or injury

- Do you feel that the content on parent-child communication reflected common challenges of parenting a child with a visible difference?
- Was there anything that you felt was missing?
- Do you feel that story telling exercise was easy to understand?
- Did you try this exercise yourself? If so, how did you find it?

Other people’s reactions

- Do you feel that the content about “other people’s reactions” reflected common challenges of parenting a child with a visible difference?
- Was there anything that you felt was missing?
- Did you feel that the content about “telling other people about your child’s condition or injury” reflected common challenges of parenting a child with a visible difference?
- Was there anything that you felt was missing?
- Did you feel that the template to tell others about your child’s condition or injury was easy to understand?
- Did you try this exercise yourself? If so, how did you find it?

Staying informed

- Do you feel that the content about “staying informed” reflected common challenges of parenting a child with a visible difference?
- Was there anything you felt was missing?
- Did you feel that the Q and A template was easy to understand?
- Did you use this template? If so, how did you find it?

Finding the right support

- Did you feel that the content about “finding the right support” reflected common challenges of parenting a child with a visible difference?
- Was there anything you felt was missing?
- Did you feel that the link list of support resources was helpful?
- Was there anything you felt was missing from these resources?

Wrapping up

- Can you see yourself or others in your family using these materials?
- Is there any other feedback you would like to share

Public involvement template analysis with exemplar quotes

The following quotes have been anonymised and included with participant consent.

Theme 1: Meeting the needs of parents

1.1. Relevant and contemporary issues

"I felt there's nothing out there really, well I've never come across anything, for parents, there's always the information you need about the condition or that type of thing but there's never that self-help for yourself, for your thoughts and feelings and how to manage them, whether it's the right thoughts I'm having, is it bad? Is it good? And so, I really felt like it was the first time something really hit the spot. I could've done with that sort of thing years ago, to be fair it's still quite helpful now with the different ways and coping strategies." *Amelia, mother of son*

1.1.1. Feeling heard and represented

> *"It made me feel quite satisfied that was across the board, because you've written this research in all different physical appearances and it fitted. So, if I was a new parent I would think 'yes these are the kind of thoughts I'd been having, so it's going to be useful'" Charlie, mother of daughter*

1.1.2. Meaningful and relevant exercises

> *"I think the writing technique is very good because it gets it out of your head, because as long as you're harbouring these thoughts in your head, you can't move on." Freja, mother of son*

> *"That bit with the professionals because it's useful, the template, to go into an appointment with the practical things like that there, absolutely fab, because that's what you're looking for, that's what I'm looking for."Katherine, mother of son*

1.2. Flexibility of materials

"If they come across it as completely new parents and give them a starting point to get them thinking about it." Vicki, mother of daughter

"Yeah, just other really good techniques, you can pick and choose and do what's best for you and *what works"Charlie, mother of daughter*

"I thought it was a really good way for different people so if you're stuck you've got something there that you can just purely use, but if not, you can use bits of it as and when and adapt it as you like.*"Amelia, mother of son*

"It normalises it doesn't it. In that yes, they are unique but if they want to be the same as everyone *else well actually they are. It's only one thing that's different"* Charlie, mother of daughter

"It just shows that it doesn't define them, that's what I got from it. They still have a favourite colour, *they're a child."Amelia, mother of son*

1.3. More content needed

1.3.1. The role of school

> *"Being aware of this before your child starts or linking in with the pastoral team, asking what support is available. Getting to the point where you're telling an adult or telling school, sometimes it's a bit like 'what comes next?'".* Katherine, mother of son.
>
> *"I think that's one of the things lots of parents I speak to are so anxious about, whether it's starting school or whether it's the transition with school. I guess even like just alluding to*

supporting parents to then have those preparatory conversations with somebody in the school environment" Ida, Clinical Psychologist.

1.3.2. Advocacy for child

"If you're going to teach the child to ask or answer questions, they have to be given the opportunity. If the health professional is looking at you, to answer a question then you look at the child" Freja, mother of son.

"The amount of kids who I've seen who literally have no idea why they are coming to the hospital and then they're in an appointment where they're being talked about…I am aware that there are lots of services that kids are going to where really complex stuff is being discussed and unfortunately sometimes that's not being thought about how the child is feeling. They've got no idea that they might be having a complex surgical procedure and parents just haven't or have been too worried I think to kind of prepare them for that." Ida, Clinical Psychologist

1.3.3. Parent knowledge

"It's very difficult to talk about these things when half of the time parents don't understand any of it. I think that causes lots of blockages. How are you supposed to talk about something if you don't even understand it?I do think information is power. Understanding something unlocks something. Then you can make these connections more easily and respond more spontaneously, more quickly."Keira, CEO and Founder of charitable organisation

Theme 2: Communicating complex concepts

2.1. Abstract concepts

"I think you've broken it down so that it's clear enough and you're not scaring someone with this theoretical model, it's just breaking it down for them." Kiera, CEO and founder of charitable organisation

2.2. The importance of examples

"The examples around that are very, very good. I love seeing examples like that. When I see examples, I can then imagine how to do it myself, it's much easier for us to… I think that the examples are a very good way to teach parents how they can do it and then make their own version."Amelia, mother of son

"I do think that actually seeing people's circumstances who may be very different and sharing similar challenges is actually really, really valuable because it makes me feel less alone and you realise 'there are other parents who are struggling with this as well'. Kiera, CEO and founder of charitable organisation.

2.3. The role of metaphors

"I generally used metaphors myself to describe things so I like that concept, because I do think difficult things are more easily explained with a metaphor that you can understand because you can relate it to the other thing" Charlie, mother of daughter

"The tree analogy, I think that was a really nice analogy and a really nice way of structuring the beginning of that conversation and giving clear guidance as to what the toolkit is all about" Alisha, audiologist working with craniofacial conditions

"I was pushing the beachball down! It's certainly one you can give that analogy to your children. Think, you've got some water and you're trying to push a ball down, it's one you can use. You can do it together physically as well." Vicki, mother of daughter

2.4. Difficulties with comprehension

"I think when it says some passengers are feelings like anxiety and I was confused, are they feeling anxiety and then I realised no they are anxiety, so just getting my head around that bit and then I realised what they represented" Amelia, mother of son

"I do think it's true that sometimes you can get caught up in trying to kind of explain that concept. One of the things I talk about is just it's really natural for us to experience negative thoughts and that our brains have kind of..as we've developed language and become more complex, that unfortunately that has some downsides, so you could almost shorten that section and kind of help people to understand that what they're experiencing is normal" Ida, Clinical Psychologist

Theme 3: Getting parents on board

3.1. Building trust with parents

3.1.1. Parent anxiety

> *"I think it takes a lot of bravery and courage and so it did bring a little bit of anxiety up." Katherine, mother of son*
>
> *"I resonate with that, because it fits in with the fact that if you don't feel brave enough or courageous enough to saying those things back or it might not be appropriate then at least that's another kind of compassionate action that you can give without being verbal." Vicki, mother of daughter*

3.1.2. Human connection

> *"Getting parents on board and helping them to think 'Yes, the person who has written this gets what I'm worrying about, thinking, and feeling about'" Ida, Clinical Psychologist*

3.2. Tapping into parent motivation

"I think what you're doing is probably, a lot of parents might feel they need that justification to take time for themselves. They're going out looking to help the children but the whole point is help yourself to help them" Katherine, mother of son

It might be the way that you phrase it in terms of putting the fact that it's ok to seek support for yourself because by helping yourself you can therefore, you're in a better position to help your child." Vicki, mother of daughter

"You've actually put "supporting your child with an appearance-affecting condition or injury ". You've actually spelt out what it's for." Vicki, mother of daughter

Theme 4: Accessibility of format

4.1. Language

4.1.1. Tone of writing

> *"I felt like it was light-hearted, it made me laugh and chuckle in my head. It felt less heavy and intense" Amelia, mother of son*
>
> *"You've really set it out in not a patronising or condescending way, it's a 'this is what we've found, this is how we think it will work'" Alisha, audiologist working with craniofacial conditions*

4.1.2. Changes to language

> *"I wasn't used to the word, "sit with any difficult thoughts and feelings" and the word sit, and I used the word cope or deal with and so until I got to page 30 where I saw it again and it read ok but initially to sit with, I don't know, to me it's deal with" Amelia, mother of son*
>
> *"The first paragraph it ends with 'how we show up for our families', I think that could be quite antagonistic for some parents reading. I would get the message across in a different way." Ida, Clinical Psychologist*

4.2. Layout

4.2.1. Visual presentation

“You’ve used the colour blocking as ways to pick out example whereas this was sort of all in together so I think the bit where you’re displaying it, the actual technique, could be made to be different in some ways. In a text box, or a picture, or whatever. If there was a way to represent it differently so it stands out.”Katherine, mother of son

“I think that’s a really good idea to actually have a little box that says ‘research shows us. I think you will find then that you are just bullet pointing the main things and then can kind of focus on what you actually want to tell parents. What are the messages you want to get across?... Yeah, and they can run through the booklet, you can have one colour that is about the research, and then use another colour about something else” Ida, Clinical Psychologist

“Yeah, I like the images, it’s something different rather than usual stock images” Amelia, mother of son

4.2.2. Interactivity

“But having a link to listen to, to give you feedback with a human voice is very valuable to that experience, because you can do it in real time then, while you’re listening to the voice, rather than reading it out of the book and trying to work out what you got to do.” Charlie, mother of daughter

“I really like this page because I think it sort of, it’s more interactive and it gives you a task to do. I like it visually; it has a change from the text.”Katherine, mother of son

4.2.3. “There’s no getting away from the fact it

“There’s no getting away from the fact it is long, and it’s long for a reason because there’s lots of important stuff in there.”Ida, Clinical Psychologist

"You've got your contents page at the beginning and then what I would do is probably skim and pick things out, and if something is really obvious you can go back and find it if it was in a bubble. I think that works really well to break that text up a bit more. But also, how you explain, sort of have mini contents, you've got section 1 to say 'this is gonna give', you know the key things so if you wanna go back over it again if you're like 'where's that breathing technique?" Katherine, mother of son

Public involvement workshop table of changes

Intervention section	Feedback	Proposed changes	Changes to be made	Rationale	Corresponding theme
Introduction: Why focus on supporting parents?	"…. I think if you could actually have a very short, very visual as you did, summary of what you're basing it on saying 'we know that this is what parents want and need" (Professional) "Why focus on the support needs of parents', I think there is that acknowledgment of parenting in general being challenging and then parenting a child who has a visible difference, the additional challenges, but I almost feel like that could be more of your headline grabber to help people to think 'Ok why is this for me?'. To help people highlight that this person understands where I am coming from and what I'm going through and what the concerns are that are going round my head." (Professional) "I thought it did, it's really obvious parents want to help support their kids, but I think sometimes how to best do that isn't quite clear. You've really set it out in not a patronising or condescending way, it's a 'this is what we've found, this is how we think it will work'" (Professional)	Reorganise introduction section to grab parents' attention and encourage them to read on	Restructure the introduction section to have a summary section at the top to introduce what the toolkit will cover.	Although parents felt that the introduction flowed well, they already knew what to expect and professional see a much wider range of parents and families who might need more encouragement to buy into the toolkit and read on. Add any evidence from the literature – take from text above	"Getting parents on board"
Why do we have difficult thoughts and feelings?	"When you actually go back to early humans, and predicting and spotting, I think that's where you lost me a little bit. I'm not sure whether the message on page 8 and 9 can be said in a different, in a totally different way. I think you risk losing people on that one." (Professional) I think that it's interesting hearing P1's perspective on that as well because I think it is so ACT'y. But, it's one of the things that I find, like if I'm doing ACT with people I kind of gloss over it…I talk about is just having 'tricky	Shorten this section. Add a section to highlight that this is the	Reduce this section and add some signposting to communicate to parents that this is	The parents who took part liked and related to this section, so it is important to retain some of the content. However, professionals felt that some parents might get lost here and lose motivation to	"Getting parents on board" Communicating complex concepts

	brains' in the way that we develop, that actually it's really natural for us to experience negative thoughts and that our brains have kind of… As we've developed language and become more complex, that unfortunately that has some downsides, so you could almost shorten that section and kind of help people to understand that what they're experiencing is normal and is… almost just move a bit quicker on from it I guess." "I liked the background about our ancestors and sort of then going into today's life, that was fine for me" (Parent) "I found it very interesting, I think it's interesting for people to read this sort of thing that they can understand as well, I have got nature in me, and I don't think people realise how much people are constantly scanning with their eyes for danger. (Parent	introduction (setting the scene), and that examples and exercises (more concrete content) will come later. Parents related to the evolutionary explanation to difficult thoughts and feelings.	setting the scene to better understand their own thoughts and feelings and how to support their children.	continue. The parents involved were mostly highly educated, so the lengthier description might not be accessible to parents from all wider range of backgrounds.	
ACT metaphors and concepts	"But I think you've used lots of great analogies, but I was trying to think about it from someone who hasn't heard ACT before and thinking maybe it flips too quickly between the two. Obviously, they are really great metaphors and that's what ACT is about, I was trying to think that it goes from somebody who's not had that background information or doesn't have a therapist who's talking through those concepts with them, then maybe actually just trying to lessen some of the information on certain metaphors and stick with one a bit more?" (Professional) "Yeah, I think so too, I like the ball one I think it just gives people a way to understand, it just gives a way to understand and think about why they might be feeling like that and result of doing that. Like the ball will just keep popping up" (Parent) "I think when it says some passengers are feelings like anxiety and I was confused, are they feeling anxiety and then I realised no there are anxiety, so just getting my head around that bit and then I realised what they represented" (Parent)	Too many metaphors, perhaps use fewer or combine into similar scenarios to reduce too many different metaphors. Increase clarity of writing and more diverse samples in some metaphors.	Keep all the metaphors but ensure that the wording and instructions are clear for each. Change wording passengers on the bus of this section to increase clarity.	By keeping the range of metaphors, it gives parents choice to explore different metaphors and find the one that works for them. Checking for clarity will help with comprehension.	Communicating complex concepts.

	“So, putting my negative thoughts at the back of the bus, where they didn’t bother me so much but then I didn’t have in the picture any pleasant thoughts, I then had to create that myself rather than being guided” (Parent)		Add positive thoughts to passengers on a bus.		
Getting hooked by thoughts and feelings	“My instinct was to say ‘getting paralysed’ by thoughts and feelings. For me the word ‘hooked’ I associated with something different, I associate getting hooked by something as getting excited by it, whereas you are talking about the anchor and it’s weighing you down, the anchor has also two meanings but it’s weighing you down, slowing you down, making you unable to engage. I wasn’t sure about the words ‘hooked’” (Professional)	Suggested to change the language.	Keep getting hooked phrase but check that the meaning is clearly explained in the text.	Only one professional expressed this opinion and all parents understood and related to the concept of “getting hooked”.	Communicating complex concepts Accessibility of format
Breathing technique	“I would say on page, the one with the hands, I don’t know if you should make the bits in blue also bullet points? I don’t know whether it’s worth putting numbers for that section, so it’s really sort of clear?” (Professional) it’s trying to explain that focusing on a breathing technique isn’t easy, but people often think that they can’t do it therefore they don’t, whereas it is something that you practice and be aware of what happens when you do it. Just making people aware that it is not easy and you will find it useful if you can stick with it (Parent) I would probably add in about instead of the whole hands, but another thing that is recommended is to lay your hands on your tummy so you can feel the rise and fall. If it’s a bit too much concentration to think about the hands opening and closing. (Parent)	Add numbers to breathing exercise to add clarity to instructions. Add a sentence about the importance of sustained practice of techniques and self-compassion during practice. Add alternatives to hand movements.	Add numbers and check clarity of exercise. Add sentence about how breathing exercises require practice. Provide choice if following the hand movements is too complex.	Public involvement members felt that some of the instructions were unclear in the breathing exercise and that having an audio recording option to follow the exercise along in real time would be beneficial. Help motivate parents if they are finding the exercise difficult and encourage self-compassion. Increase accessibility of the exercise and prevent parents becoming frustrated if they find the exercise challenging.	Communicating complex concepts Accessibility of materials.

Grounding techniques	I do find it a useful thing, sometimes I find 4 or 5 things difficult to find. I don't know if it's ok to say 3. I think 2 probably isn't enough. Especially if I'm doing it more than once and I want different things. But I do find it useful, again a bit like focusing on the breathing it actually is a way of focussing away from your overwhelming thoughts and feelings and bringing the attention back on something else more useful (Parent)	Limiting the number of grounding activity senses to no more than 3.	Keeping the number of senses at 3.	Reduce complexity of the exercise to increase accessibility.	Accessibility of materials
What are values?	"I don't know if it's a really text heavy page, I don't know if in the booklet it maybe a double spread. So, there's some on that side, so it's still open and they can still see all of it, but they can spread it out. But especially the bottom where you want them to write something, but I don't envision the bubbles being big enough. I suppose you can move some over of just make those bottom three bigger bubbles, so just have a page of just big bubbles." (Professional) "Concept definitely and you've made it clear that it's come from the research, etc. Oh, have you actually? I don't know whether you could says 'these values have come from my research', I don't know whether this is important to say?" (Professional) Pt 1: I think it's a work in progress as well and I don't know whether you wanted to add that in anywhere, that if you do this exercise, it's not a complete document on the day, because things that didn't occur to you that are really important will pop up in thoughts or in conversations at a later date. You might think I want to add that to it. It would be nice to know that this is something to keep coming back to Pt 2: Or they might change as they go along, as life goes on things happen and they might change. I think to include that is a good idea (Parents)	Change formatting to make exercise a bit clearer to read and use Add links to research related to values exercises Add that values exercise can change and evolve over time.	Make changes to value identification exercise to enhance clarity and reduce possible confusion in parent users. Possibly using visuals to help in this process. Link the concept of values to the research findings.	Values are a complex concept and this seemed to be the area parent public involvement members struggled the most.	Communicating complex concepts Accessibility of materials
Talking about your child's condition or injury	"It's very difficult to talk about these things when half of the time parents don't understand any of it. I think that causes lots of blockages. How are you supposed to talk about something if you don't even understand it?" (Professional)	Add a sentence about the importance of knowledge.	Add reference to gaining knowledge about child's condition or injury here and in the	Important to empower parents to seek information about their child's condition/injury, so they can pass this knowledge onto children. But also important to remind parents to be self-	Meeting support needs

			introduction to encourage parents to engage. Also acknowledge that is not possible to know everything.	compassionate in their approach to this.	
Telling other people about your child's condition or injury	"I think that could potentially be really good. You might not even need a really long section, it might just be like 'here are some things to think about'. You know, whether to arrange to have a meeting with school, to have those conversations with a class teacher or a headteacher or whoever it is around what the child might want to be shared or discussed or how things are talked about in the family. Then like you say, also then giving like a link to that would kind of be full circle, I guess. It would be nice." (Professional)	Add link to teachers guide Highlight the transition to school more specifically and things parents can do to support their child going to/moving schools.	Include a more explicit section on schools.	School transition is a salient issue for parents and is often mentioned in research. Existing condition-specific visible difference research suggests parent concerns related to school can be a considerable challenge (Christensen et al., 2017).	Meeting support needs.
Staying informed	"So I think a little section, you can do it gently, 'preparing for seeing your doctor'. I think things like informed consent is a really important consent that I learned afterwards, too late, I hadn't asked my child for consent. Why would you never tell me about this? You don't know that word [consent] when you are parent, but it is so important" (Professional) "I don't know if it's worth doing it just above that 'clarify the points – make sure before you leave you can ask for a summary, check your understanding'" (Professional) It think to add something in there to remind parents to definitely look up their options and don't always take it on face value, there might be other options, seek support elsewhere and speak to others in similar situations (Parents)	Add a section on preparing children for appointments, advocacy and informed consent. Add section on checking understanding with HCPs before parents	Add these sections Reminding parents that they are the expert of their child.	Preparing children and advocating for children in a healthcare setting is an important issue and can help to teach the child how to respond in these environments as they grow up. Existing paediatric chronic illness literature has stated the importance of including children and young people in decision	Meeting support needs

		leave appointments Adding that parents are often very knowledgeable and the experts of their child’s condition.		making involvement (Miller et al., 2018).	
Finding the right support	“Maybe you say something about like ‘These are trusted sites that we recommend you would use rather than going down that Google blackhole’ or something. Just expand on that a little bit just to make it really clear that there is so much information online, how do you choose the best information?” (Professional) Include a summary of the support links included, like please see condition-specific overleaf to signpost people (Parent)	Add rationale to online support section. Signposting to condition-specific information	Clarify that these websites are trusted and contain reliable information and sources of support.	Reassure parents that the information provided within the materials can be trusted.	Getting parents on board
General formatting and language	“I hadn’t thought of that, but I think that’s a really good idea to actually have a little box that says ‘research shows us’. I think you will find then that you are just bullet pointing the main things and then can kind of focus on what you actually want to tell parents. (Professional) “one type of document then maybe there would be a summary document, like a five-page pamphlet that summarises it and that would overcome the problem of it being too wordy but you then, have another document! Because sometimes you really want that amount of information as a parent but sometimes, maybe in the first instance you might want something really quick to scan through to then nowhere to get all this information from. Almost like a summary pamphlet that promotes it”	Use textboxes and colour to highlight information from the research. Addition of a summary document	Change this formatting. Consider as a possibility post-PhD to increase accessibility of the information in the document for those who do not wish to	Increase readability and reduce burden on parents.	Accessibility of materials

	At the outset you mention parents and carers, I work in the NHS with children in care, so every time you mention parents, you possibly need to mention 'and carers' just for consistency. The focus is absolutely brilliant, and it sets the scene, but just to be aware that you start off saying 'parents and carers', you need to follow it through. (Parent)	Ensure consistency in language	read the entire toolkit. Check for consistency.		

Appendix L: Study three materials

Acceptability survey:

The first thing you will need to do is create a unique participant code.

As your answers are anonymous this code will be used to identify your response if you wish to withdraw your data.

First, please enter the number of the day you were born e.g., if you were born on the 11th of January, you would enter 11.

Now, please enter the last letter of your first name e.g., if your name is Alice, you would enter E.

Finally, please enter the first letter of the name of the place you were born e.g., if you were born in London, you would enter L.

Thank you for creating your unique participant code.

I would now like to ask you a few questions about yourself. Your answers will help me to understand a bit about your background and your family.

How old are you?

Which gender do you identify with?

Female
Male
Non-binary
Prefer to self-describe
Prefer not to say

How would you describe your ethnic background?

Asian/Asian British (Bangladeshi, Chinese, Indian, Pakistani, Any other Asian background)

Black/African/Caribbean/Black British (Any other Black/African/Caribbean background)

Mixed/Multiple ethnic groups (White and Black African/White and Asian/White and Caribbean/another other mixed/multiple ethnic background)

White (British/English/European/Gypsy or Irish Traveller/Irish/Norther Irish/Scottish/Welsh/Any other White background)

Other ethnic groups (Arab/Any other ethnic group)

How would you describe your marital status?

Single
Married/Civil partnership
Divorced
Separated
Cohabiting
Other (please specify)

☐

Who do you live at home?

☐

How many children do you have?

What is your relationship to your child with an appearance-affecting condition or injury?

I would like to ask you a few more questions about your child, so that I can better understand your experiences as a parent or carer.

What condition or injury does your child have?

Please describe how the condition or injury affects their appearance?

How old is your child?

What is their gender?

Female
Male
Non-binary
Prefer to self-describe
Prefer not to say

This series of questions will be about how easy the parenting toolkit is to understand.

Please read the statements below and select the option that feels most relevant to your experiencing when using the toolkit.

	Strongly disagree	Disagree	Neither agree nor disagree	Agree	Strongly agree
I understood the information in the parenting toolkit.					
I understood the exercises in the parenting toolkit.					
I understood the metaphors in the parenting toolkit (e.g., tree metaphor, passengers on a bus, beach ball etc.).					
The language used in the parenting toolkit was easy to understand.					
The images used in the parenting toolkit were easy to understand.					
The images complimented the text.					

Was there any aspect of the parenting toolkit that you found difficult to understand (e.g., confusing, or difficult language, concepts not explained well)?

Is there anything else you'd like to add about how easy or difficult the parenting toolkit was to understand?

Were there any important topics that you felt were missing?

The next series of questions are about how comfortable and confident you felt using the parenting toolkit.

Please read the statements below and select the option that feels most relevant to your experiencing when using the toolkit.

	Strongly disagree	Disagree	Neither agree nor disagree	Agree	Strongly agree
I felt confident engaging with the parenting toolkit.					
The parenting toolkit would be helpful to me and my family in the future.					
The people who contributed to parenting toolkit understand what is important to me and my family.					
I learnt something from the parenting toolkit.					
I value the information provided in the parenting toolkit.					
I trust the information provided in the parenting toolkit.					

I feel more confident about supporting my child after reading the parenting toolkit.					

Was there anything that you felt prevented you from using the parenting toolkit?

Was there anything in the parenting toolkit that you didn't find helpful?

The next series of questions are about how it felt to use the parenting toolkit.

Please read the statements below and select the option that feels most relevant to your experiencing when using the toolkit.

	Strongly disagree	Disagree	Neither agree nor disagree	Agree	Strongly agree

I enjoyed reading and using the parenting toolkit.					
The content of the parenting toolkit felt relevant to me.					
The content of the parenting toolkit is in line with my family values.					

Is there anything you would like to add about your experience of reading and using the parenting toolkit?

The next series of questions will ask you to think about your overall experience of using the parenting toolkit.

Please read the statements below and select the option that feels most relevant to your experience when using the toolkit.

	Very poor	Poor	Neither good nor poor	Good	Very good	Don't know
Overall, how was your experience of the parenting toolkit?						

	Extremely unlikely	Unlikely	Neither likely or unlikely	Likely	Extremely likely	Don't know
How likely are you to recommend the parenting toolkit to friends or family if they need support?						

Thinking about your responses to these last two questions. Please can you say a bit about why you feel this way.

Is there anything about the parenting toolkit that you think could be improved?

These final few questions will ask you to think about which sections of the parenting toolkit you used and what you found to be most or least helpful.

Approximately how long did you spend reading and using the parenting toolkit?

Did you think the parenting toolkit was:

1) Too long
2) Too short
3) Just right

Is there anything else you'd like to say about the length of the parenting toolkit?

Which sections of the parenting toolkit did you access?

1) Introduction (why support parents?)
2) What are difficult thoughts and feelings?
3) Helper skills
4) Being present
5) Knowing your values
6) Talking about your child's condition or injury
7) Other people's reactions
8) Staying informed
9) Finding the right support

Please use the boxes below to rank the parenting toolkit sections from "Least helpful" to "Most helpful" (using ranking exercise software on Qualtrics)

- Introduction (why support parents?)
- What are difficult thoughts and feelings?
- Helper skills
- Being present
- Knowing your values
- Talking about your child's condition or injury
- Other people's reactions
- Staying informed
- Finding the right support

Did you try any of the exercises in the parenting toolkit?

1) Yes
2) No

If so which ones?

Which exercises do you feel would be most helpful for you and your family?

If the parenting toolkit were available to you and your family how often do you think you would use them?

Is there anything you'd like to add about your experience of the different sections of the parenting toolkit?

Is there any other feedback that you would like to share?

That is the end of the survey, thank you so much for taking part!

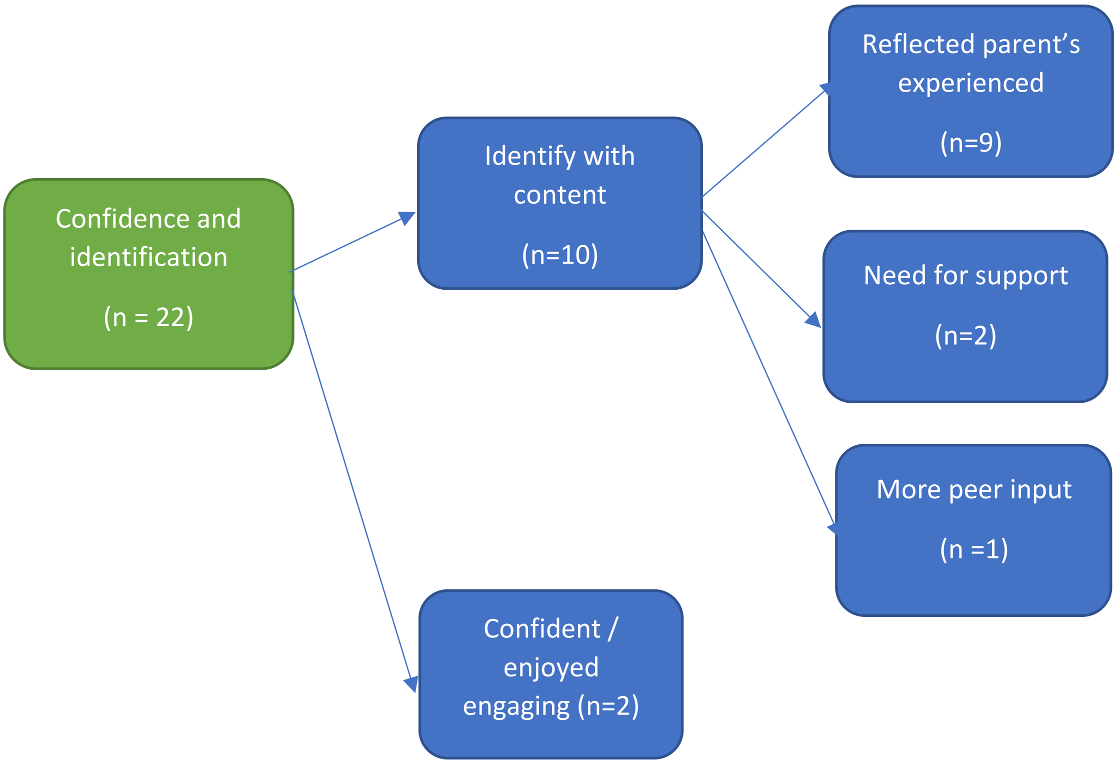
I really appreciate you giving your time to support this research.

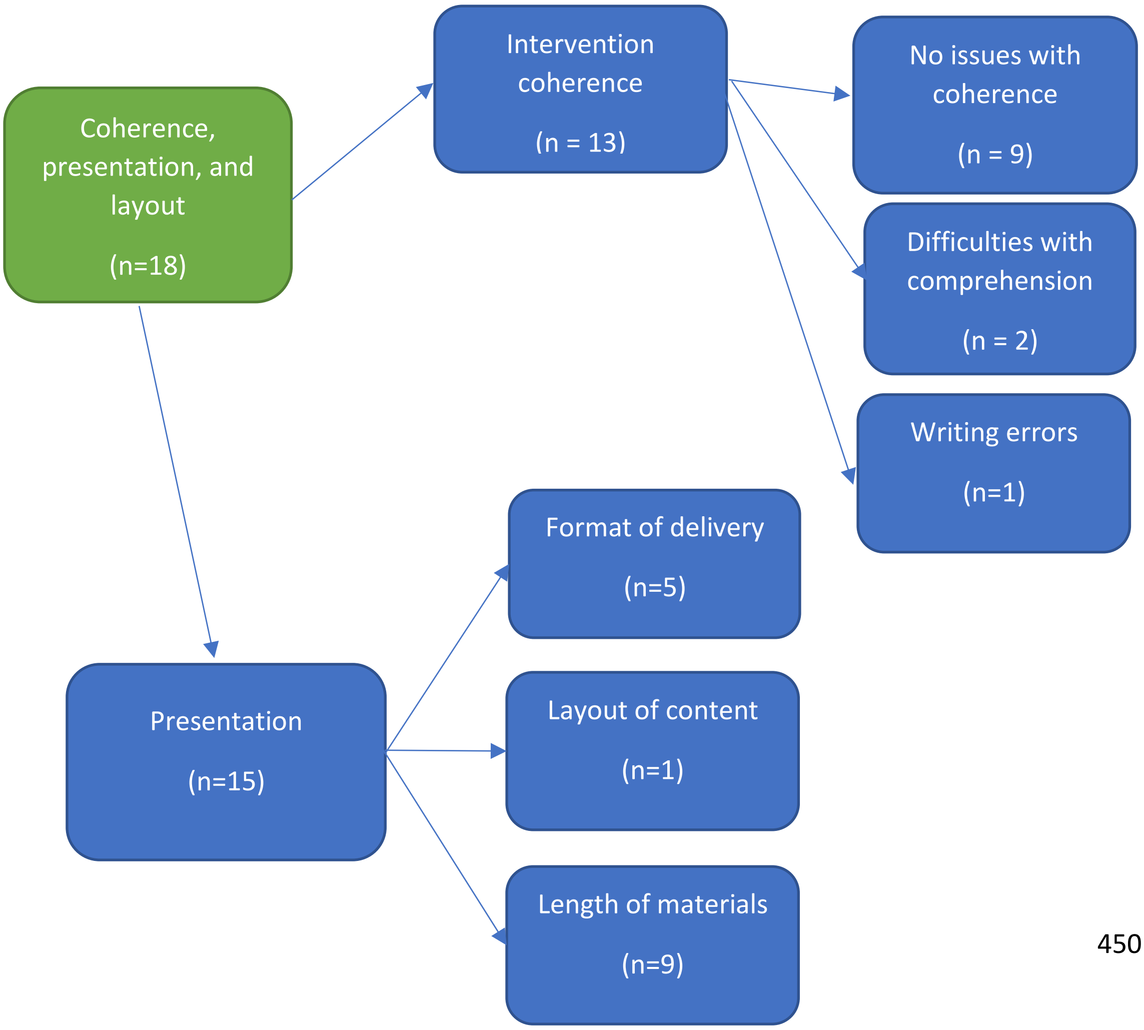
Your responses will help us to better understand how best to provide support to parents and carers of a child with an appearance-affecting condition or injury.

If you would like to be entered into the prize draw to win a £50 Amazon voucher, please enter your email address below (this email address will be stored separately to your survey responses).

If you would like to receive an email update about the next steps of this research, please provide an email address below (this email address will be stored separately to your survey responses).

Study 3 content analysis categorisation matrix





Study 3 content analysis exemplar quotes

Theme	Sub-theme	Exemplar quote
Confidence and identification	Identify with content	“It was well structured and was definitely in tune to the thoughts that parents may have” “It was good to realise that some of the methods we've used are suggested like openly talking about things in a natural way to 'normalise' her condition and talk about how other people might see her as different.”
	Confident/ Enjoyed engaging	“This would be a favourite, long time resource for us” “It was very informative. I enjoyed looking and trying out the exercises. It made me think of things in a different way.”
Coherence, presentation, and layout	Intervention coherence	“I understood the whole toolkit very good resource.” “I thought it was completely at the right level - no jargon or wording I didn't understand and also not condescending.” “At the very beginning the document talks about 'carrying' things for my child. I think this meant carrying emotions, but I struggled a bit with this language and it was a little off-putting so close to the beginning of the document”
	Presentation	“Layout good - not overwhelmingly volume of text on each page.” “It was a little long but that is only a very minor thing.” “It’s very long, I had to review in multiples session, but I got so much out of it!” “I would love a paper copy if and when available please.”

Appendix M:

This appendix has been redacted.