

**Psychological adjustment in cleft lip and/or palate:
Outcomes, methods and neglected groups**

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Thesis for the degree of Doctorate of Philosophy by published work (DPhil)

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Abstract

To date, research examining psychological adjustment to cleft lip and/or palate (CL/P) has produced conflicting findings. As a consequence, large gaps in our understanding of CL/P remain, ultimately impacting the ability to implement and evaluate psychological support for this population. The work presented in this DPhil discusses the conceptual and methodological challenges associated with this field in accordance with three key underlying themes: outcomes, methods and neglected groups, in an attempt to explain why these conflicting findings occur and to offer an alternative perspective. Specifically, this doctoral thesis proposes the need for an approach which is holistic rather than narrow, inclusive rather than exclusive, normalising rather than pathologising, appreciative of the patient perspective and encompassing of patient strengths and positive growth.

Author statement

From a young age I knew I wanted a career in Psychology, but it wasn't until I began volunteering at the Centre for Appearance Research (CAR) at the University of the West of England (UWE) while studying for my undergraduate degree that I first considered academic research. Initially I was unconvinced, in my mind linking research with dull statistics and office work. Over time however, I discovered that not only is research diverse, exciting and involves plenty of patient contact, but that through research there is the potential to make much larger changes to people's lives than is arguably possible on a one-to-one clinical basis. It also allowed me the freedom and flexibility to work independently, the space to be creative, and the opportunity to indulge my love of writing. Progressively, I was asked to present my work at a range of multidisciplinary national and international conferences. I began to build up networks of stakeholders, from other psychologists to surgeons, charities and funding bodies. During this time I also completed two Masters degrees; the first in Research Methods in Psychology and the second in Health Psychology. I felt challenged every day and I was learning at a fast pace.

A second wave of doubt arose when I realised that all of my work had started to become focused on a single field: cleft lip and/or palate (CL/P). Not only did I not want to be pigeonholed into one area of work, but I couldn't see how my thirst for knowledge and clinical application could be satisfied by a single condition. However, I came to learn that CL/P was a lifelong, complex condition, involving not just the individual but the whole family, as well as an array of different medical disciplines and treatments. I discovered psychological adjustment to CL/P to be multifaceted and to fluctuate over time according to ever-changing situations and life events. I became fascinated with the question of why one person could seemingly adjust to the condition so well, while another person exhibited ongoing difficulties throughout their life. What was it that made these two people's experiences so different and how could we attempt to implement this knowledge into psychological intervention?

In trying to find answers to these ambitious questions, I found the existing literature to be marred by a multitude of methodological limitations. Not only was everyone using different measures, approaches and terminology, but no one

seemed to agree upon what it is that constitutes a positive psychological outcome. While understanding there is a case to be made for the necessity of psychological intervention, I was surprised by the ongoing focus on deficits and problems, with very little work being carried out on resilience and personal growth. I also found that key groups of patients were being excluded from research samples, and I was frustrated by the tendency for authors to largely neglect any additional challenges (such as comorbid conditions) and individual variation. Finally, I was disappointed by the lack of evidence of the benefits of psychological support and intervention, and the lack of coherence in the interventions offered. Psychologists are an expensive commodity, and as such need to be able to measure, evaluate and demonstrate their value and impact. It was the identification of these gaps in the knowledge which drove my curiosity in this area, and it is these discoveries which form the key themes integrated into this doctoral thesis.

Throughout the course of preparing for this DPhil, I have grown both professionally and personally. I have learned a great deal about my strengths and the areas which would benefit from further development, as well as about the research field itself and the differing methodological approaches that can be utilised. I have felt incredibly privileged and inspired to be the recipient of participants' past experiences and hopes for the future. I have also been remarkably fortunate to meet and collaborate with like-minded clinicians and researchers with similar ideas, values and ambitions for the field.

Today, I feel in my element. On a daily basis, I am responsible for the psychological component of the largest cleft lip and palate research programme in the world. Aside from seeing my ideas implemented into ongoing research and practice, I feel my hard work is recognised and rewarded by my colleagues and by the university. My work is driven by original ideas and personal values, including the necessity to conduct research which is creative, reflective, well-designed and well-communicated, and which leads to new knowledge for the benefit of the field and the recipients of CL/P services, both in the UK and abroad. I hold no doubts that this career poses many stimulating opportunities, and am incredibly excited to be a part of the future of this field.

Declaration of authorship

| Publication | Role |
|---|--|
| Rumsey, N. and Stock, N.M. (2013) Living with a cleft: Psychological challenges, support and intervention. In: Berkowitz, S., ed. (2013) <i>Cleft Lip and Palate: Diagnosis and Management, 3rd edition</i> . Berlin Heidelberg: Springer-Verlag; pp. 907-915. | <ul style="list-style-type: none"> • Completed comprehensive literature review • Collaboratively drew conclusions and corresponding implications • Contributed to the writing and editing of the book chapter • Disseminated the findings |
| Feragen, K.J.B., Stock, N.M. and Rumsey, N. (2014) Towards a reconsideration of inclusion and exclusion criteria in cleft lip and palate: Implications for Psychological research. <i>Cleft Palate-Craniofacial Journal</i> , 51 (5), pp. 570-579. | <ul style="list-style-type: none"> • Contributed to the overall concept and design of the project • Conducted a literature search • Contributed to the interpretation of the statistical analyses • Collaboratively drew conclusions and corresponding implications • Contributed to the writing and editing of the paper • Contributed to the dissemination of the findings |
| Feragen, K.J.B. and Stock, N.M. (2014) When there is more than a cleft: Psychological adjustment when a cleft is associated with an additional condition. <i>Cleft Palate-Craniofacial Journal</i> . 51(1), pp. 5-14. | <ul style="list-style-type: none"> • Contributed to the overall concept and design of the project • Conducted a literature search • Contributed to the interpretation of the statistical analyses • Collaboratively drew conclusions and corresponding implications • Contributed to the writing and editing of the paper • Contributed to the dissemination of the findings |
| Stock, N.M. and Rumsey, N. (2015, e-pub 2013) Parenting a child with a cleft: The father's perspective. <i>Cleft Palate-Craniofacial Journal</i> . 52(1), pp. 31-43. | <ul style="list-style-type: none"> • Identified and developed the topic • Conducted a literature search • Designed the study with supervision • Collected the data • Conducted qualitative analysis • Collaboratively drew conclusions and corresponding implications • Wrote and edited the paper with supervision • Dissemination of the findings |
| Stock, N.M. , Feragen, K.J.B. and Rumsey, N. (e-pub ahead of print, 2014) "It doesn't all just stop at 18." Psychological adjustment and support needs of adults born with cleft | <ul style="list-style-type: none"> • Identified and developed the topic • Conducted a literature search • Designed the study with supervision • Collected the data • Conducted qualitative analysis • Collaboratively drew conclusions and |

| | |
|---|---|
| lip/palate. <i>Cleft Palate-Craniofacial Journal</i> , doi: http://dx.doi.org/10.1597/14-178 | <p>corresponding implications</p> <ul style="list-style-type: none"> • Wrote and edited the paper with supervision • Dissemination of the findings |
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| Stock, N.M. , Feragen, K.J.B. and Rumsey, N. (e-pub ahead of print, 2015) Adults' narratives of growing up with a cleft lip and/or palate: Factors associated with psychological adjustment. <i>Cleft Palate-Craniofacial Journal</i> , doi: http://dx.doi.org/10.1597/14-269 . | <ul style="list-style-type: none"> • Identified and developed the topic • Conducted a literature search • Designed the study with supervision • Collected the data • Conducted qualitative analysis • Collaboratively drew conclusions and corresponding implications • Wrote and edited the paper with supervision • Dissemination of the findings |

I confirm that all of the work presented in this doctoral thesis, including the chosen publications and accompanying commentary (except where stated) is the original work of the author.

I confirm that none of the published body of work included within this portfolio of selected publications has been submitted for another academic award either in this or any other institution.

I confirm that the necessary training requirements have been met (60-120 credits, of which at least 60 are at level M).

Miss Nicola Marie Stock

SEPTEMBER 2015

Introduction

“It’s not just surgery and fixing. It’s all the stuff that goes with it.”

--- Mother of two children born with cleft lip and palate.

The work presented in this doctoral thesis stems from an inherent interest in how people adjust to, and grow from, difficult situations, as well as a desire to understand human interaction and behaviour. At the Centre for Appearance Research (CAR), based at the University of the West of England (UWE), our research seeks to understand how an individual’s perception of their outward appearance can influence how they feel about themselves, how they perceive their social environment and the health behaviours in which they subsequently engage. My particular area of interest focuses on how people who are affected by disfigurement (visible difference) adjust to their condition from a holistic psychological perspective.

The following introductory section to the DPhil will discuss the importance of an often seemingly frivolous issue – the psychology of appearance – and how having an appearance which differs from the ‘ideal’, or even from the ‘norm’, can have a significant psychological impact on those affected and those around them. Specifically, this introductory section will outline the characteristics of the most common congenital craniofacial condition across the world (cleft lip and/or cleft palate; CL/P) and the delivery of treatment and support services for this population. This section will also provide an overview of the potential psychological impact of CL/P on affected individuals and their families using a lifespan perspective, as well as describe the models and theories that have been presented in an attempt to understand these impacts. Finally, this section will outline several conceptual and methodological challenges which limit the quality of the current evidence base. The aims of this DPhil will be presented, in relation to addressing these gaps and offering pragmatic suggestions for the advancement of future research and clinical practice.

The psychology of appearance

Appearance is a universal topic with relevance to all (Rumsey and Harcourt, 2005). Our outward appearance is not only unique, but plays a key part in an array of daily activities, from social encounters with others to a wide range of health behaviours. In particular, the face is central to a person's sense of identity and is the principal site of verbal and non-verbal communication (Rumsey, 2012). It is the face that is captured in photographs and that is presented on social networking sites. The face is constantly on show and is the primary focus of gaze in interpersonal encounters (Rumsey, 2012).

Throughout history, people have always been interested in making the most of their looks (Frith, 2012). In recent decades however, the emphasis we place on appearance as a society and the pressure to conform to sociocultural 'ideals' has grown alarmingly (Grogan, 2008). According to a recent survey of 77,000 adults (the largest study of appearance concerns to date; Diedrichs et al., in preparation), only 16 percent of women and 27 percent of men reported liking what they see when they look in a mirror, while 46 percent of women and 62 percent of men reported feeling ashamed of how they look. In addition to this, more than two-thirds of the women surveyed and almost half of the men reported feeling appearance-related pressure from the media. Other studies have demonstrated that girls as young as five show a preference for thinner ideal body sizes than their own (Williamson and Delin, 2001) and that body image dissatisfaction is not only common among boys but often associated with significant distress (Cohane and Pope, 2001). In adolescence, social 'belonging' is a central facet of wellbeing, with appearance becoming the prime method of evaluating this (Liossi, 2003). Further, beauty is often sold as a prerequisite for success in both the personal and professional sphere (Rumsey, 2008).

Consequently, for many children, adolescents and adults, the discrepancy between how they think they actually look and how they feel they should look is a source of significant distress. Such distress can result in poorer mental wellbeing and engagement in a variety of unhealthy behaviours, such as disordered eating, excessive exercise, the disproportionate use of cosmetics, the risky use of medication and the pursuit of cosmetic surgery (Grogan, 2008).

While the beauty 'ideal' grows increasingly unachievable for all, those who have an appearance which differs even from the 'norm' are confronted with an additional array of challenges.

Visible difference

A visible difference is the term used to describe a health condition which involves an appearance-altering component. A visible difference may be congenital, such as a birthmark or a cleft of the lip and/or palate, or be acquired later in life as a result of an injury or illness, such as a burns scar or treatment for cancer. According to the latest estimations by the charity Changing Faces, in excess of one million people living in the UK have a disfigurement to the face and/or body (Partridge and Julian, 2008).

Due to advances in medical knowledge and technology, more and more people are being saved from life-threatening conditions. Unfortunately, the result can leave aesthetic and psychological scarring. Unlike many other conditions, the 'symptoms' of conditions which affect appearance are clearly visible to others, and the individual can feel that they have lost their social privacy (Clarke, 1999). Facial and bodily function can also be affected, and the individual often has to engage with a long-term programme of multidisciplinary care.

Despite vast improvements in service provision and surgical techniques for those affected by a visible difference, psychological support to facilitate coping with the condition and its treatment still lags a long way behind. Paradoxically, psychological wellbeing is not dependent upon the objective severity or the degree of visibility to others (Moss, 2005; Ong et al., 2007; Brown et al., 2010; Feragen et al., 2010), indicating the relative importance of subjective perceptions and processes compared with objective treatment outcomes. In addition, although psychological distress can be considerable and debilitating, many individuals adjust well (ARC, 2009). Such findings suggest that adjustment to a visible difference is multifactorial, involving a complex interplay of physical, cultural, psychological and social factors (Moss, 1997; Clarke 1999; Endriga & Kapp-Simon, 1999; Thompson & Kent, 2001; Rumsey & Harcourt, 2004). Furthermore, the degree of individual variation appears to be high and adjustment likely to fluctuate over time and across situations (Rumsey and Harcourt, 2005).

Cleft lip and/or palate

A cleft (or 'split') in the lip and/or the palate (CL/P) is the most common congenital craniofacial condition, affecting approximately 1,000 live births each year in the UK (Mossey et al., 2009). While the various possible causes of CL/P remain largely untested or as yet unidentified, a disturbance in the embryologic development of the face and palate between the fifth and eleventh week of gestation may result in a cleft. The type of cleft which occurs can vary depending upon the timing and degree of the disturbance (Sperber and Sperber, 2013).

Clefts of the lip and/or palate may be classified into three main categories:

- 1) Cleft lip/alveolus (CL), unilateral (UCL) or bilateral (BCL)
- 2) Cleft lip and palate (CLP), unilateral (UCLP) or bilateral (BCLP)
- 3) Cleft palate only (CP) or submucous cleft palate (SMCP)

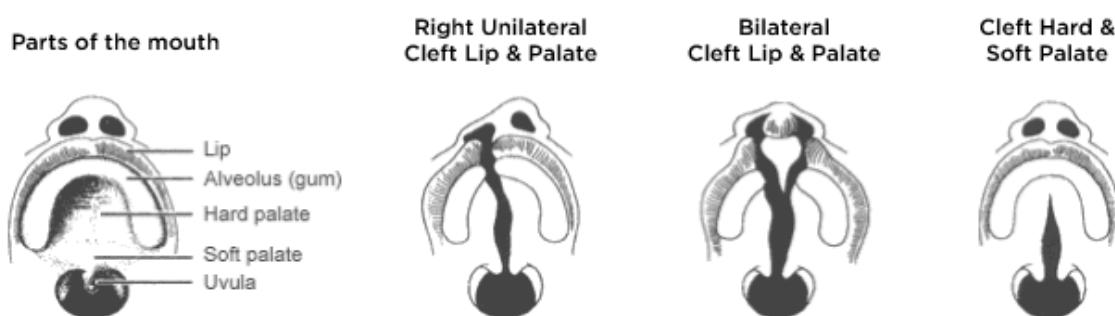


Figure 1: Classification of cleft lip and/or cleft palate. Courtesy of the Cleft Lip and Palate Association (www.clapa.com)

Although CL/P occurs most frequently as an isolated anomaly, a cleft may also be associated with a number of other congenital irregularities. In some cases, a cleft may form part of a genetic syndrome (Mossey et al., 2009).

Cleft care in the UK

Until recently, there were more than 57 active cleft teams spread unevenly across the UK. In 1998, a review of service provision commissioned by the Clinical Standards Advisory Group (CSAG) identified a total of 75 surgeons, 70 speech

and language therapists and 105 consultant orthodontists working with children affected by CL/P (Sandy et al., 1998). Unsurprisingly, these high figures indicated that few health professionals (HPs) working in the field were specialists in CL/P. Concordantly, the standards of cleft care were disappointing and the outcomes for affected children were sub-optimal.

Following this review, cleft care was centralised in the UK. Multidisciplinary cleft teams were established and located within 18 CL/P specialist centres. Expertise and resources were allocated to each centre in accordance with population needs and accessibility. Recommendations were also made for a psychologist to be incorporated into each team.

Today, most UK cleft teams are able to offer specialist nursing, reconstructive and aesthetic surgery, orthodontics, audiology, paediatrics, speech and language therapy, genetic counselling (by referral) and psychological support. In addition, supplementary information and support is offered by the Cleft Lip and Palate Association (CLAPA; www.clapa.com), the only UK-wide voluntary organisation specifically focused on helping those with, and affected by, CL/P.

The CL/P treatment pathway

In resource-rich countries, a cleft of the lip is now usually identified during the 20-week pregnancy scan. A cleft of the palate is much more difficult to detect, and is normally diagnosed after birth. In the UK, infants diagnosed with CL/P are referred to their local specialist multidisciplinary cleft team within 24 hours. Following a diagnosis of cleft, a specialist cleft nurse will provide parents with initial information about CL/P and practical feeding assistance. Families may also be invited to attend an outpatient appointment with the core members of the cleft team.

Although children with CL/P receive care which is tailored to their individual needs, a recommended timetable is provided. For families under the care of the NHS, the typical care pathway is outlined below in Figure 2. During the first six weeks, a hearing test and paediatric assessment will be performed. Surgery to repair the lip normally takes place when the child is aged three months, while the palate is repaired between the ages of six to 12 months. If the child's hearing is significantly affected (often caused by a sticky secretion in the middle ear, called

Otitis Media with Effusion, or 'glue ear'), small plastic tubes (grommets) may be inserted into the eardrum during surgery. Alternatively, or if the grommets are unsuccessful, a hearing aid may be required. Speech assessments are typically carried out at 18 months, three years and five years of age. If difficulties with the child's pronunciation and/or use of language are identified, Speech and Language Therapy may be introduced to enhance the child's speech development. Further corrective surgery may be required to reduce the airflow through the nose when speaking. Children with CL/P are also more susceptible to tooth decay, and to having missing teeth, extra teeth or teeth which are out of position. Engagement with a paediatric dentist, family dentist and/or an orthodontist may therefore be required to encourage good oral hygiene, to improve the alignment of the teeth and/or to monitor the development of the jaws and bite during growth. According to NH England guidelines (D07/S/a, 2013), children born with CL/P should have a plan in place to address any identified hearing difficulties, have dental health which is in line with other children in the region, demonstrate good quality and intelligible speech, have good maxillary growth and facial appearance and have been screened for any psychological difficulties before the age of five years.

After this, children with CL/P may require an alveolar bone graft, which aims to correct any clefting in the jaw and/or to repair the fistula between the nose and mouth. This is normally performed between the ages of eight and 11 years. Ongoing orthodontics to achieve a good appearance in the child's permanent dentition is common between the ages of 11 and 15 years. Orthognathic surgery, rhinoplasty and cleft re-repairs may be offered to patients during late adolescence and early adulthood to enhance aesthetics and/or function.

During and following completion of this pathway, regular outpatient appointments may be needed for close monitoring of the child's development and to address any difficulties. Multidisciplinary audit records are taken when the patient is aged 5, 10, 15 and 20 years of age as a means of tracking treatment progress. Until more recently, patients were typically 'discharged' from the cleft service around the age of 18 years. In the majority of cleft teams this process is now much less formal and adult patients can return to the service via a General Practitioner (GP) referral should they wish to.

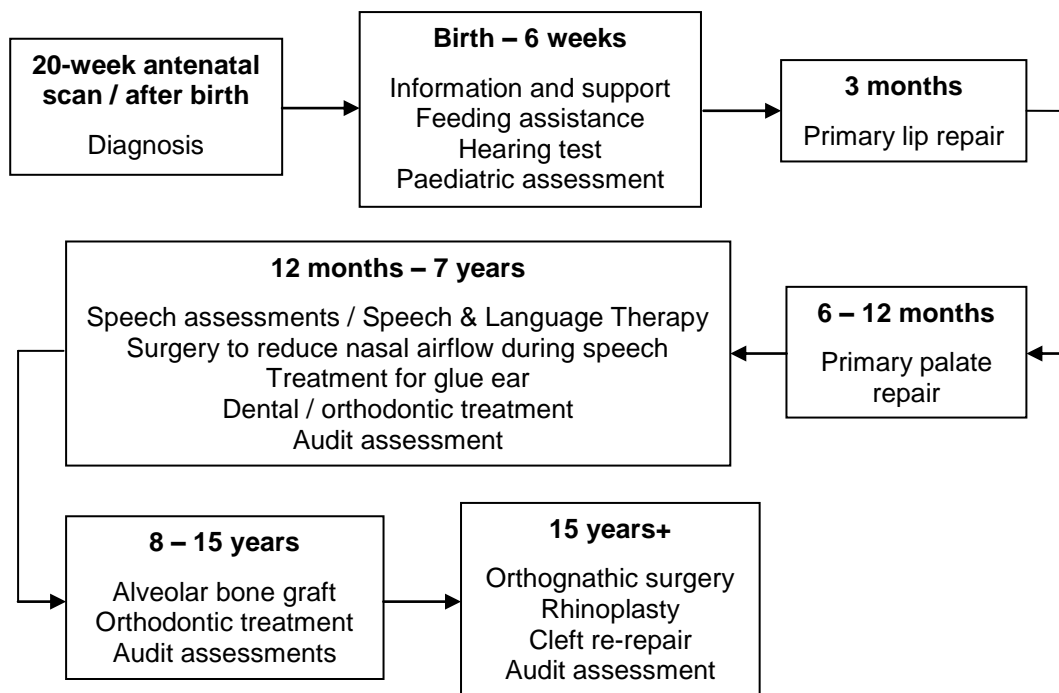


Figure 2: Typical treatment pathway for cleft lip and/or palate in the UK.

The psychological impact of CL/P

CL/P can pose a number of significant challenges for affected individuals and their families. Initially, parents may experience shock, guilt and grief upon receiving a diagnosis of CL/P in their child (Nelson et al., 2012). Questions such as ‘why has this happened to us?’ and ‘what did we do wrong?’ often go unanswered because of a lack of understanding about what causes CL/P (Williams et al., 2012). Although the quality of information and support that a family receives at this time is crucial for long-term wellbeing (Chuacharoen et al, 2009; Vanz & Ribeiro, 2011), few parents perceive non-specialist HPs to possess the knowledge, experience and expertise to provide it (Collett and Speltz, 2007; Knapke et al., 2009; Nelson et al., 2012). Following the birth of the child, feeding difficulties are often distressing and the bonding experience between the parents and infant can be disrupted (Collett and Speltz, 2007; Despars et al., 2011). The reactions of friends, family members and members of the public to the cleft also have an influence on parental wellbeing (Nelson et al., 2012). The child’s surgical repair is an extremely emotional time for the family, and parents may find it difficult to adjust to their baby’s sudden change in appearance (Nelson et al., 2012). If

genetic counselling is warranted, and an underlying genetic cause for the cleft is detected, this news can have additional implications for the whole family (Mossey et al., 2009). As the child grows older, the ongoing burden of treatment should not be underestimated. Regular outpatient appointments have the potential to impact significantly on the family's time, energy and financial situation, as well as put strain on familial relationships (Baker et al., 2009). Parents may worry about whether their child's treatments are effective, and whether their child will 'be OK' in relation to social, educational and psychological development (Nelson et al., 2012). In addition, burden of care can vary significantly across cleft teams and between countries (Semb et al., 2005).

For the child, entering into a social environment outside of the family unit can invite staring, questions and comments from others (Rumsey and Harcourt, 2005). Having an appearance which is unusual, as well as various hearing and speech complications which may interfere with communication may make it more difficult for a child with CL/P to integrate with their peers (Hearst, 2007). Seven to eight years is a peak age for teasing/bullying (Hearst, 2007). In addition, some children may experience cognitive difficulties (Richman et al., 2012; Roberts et al., 2012), which put them at risk for additional conditions such as autism, attention deficit/hyperactivity disorder (AD/HD), dyslexia, specific language impairments and developmental delay. During adolescence, appearance, romantic relationships and a sense of social 'belonging' become more important for psychological wellbeing (Liossi, 2003; Griffiths et al., 2012). Additional corrective surgeries at this age may pose a further challenge for adolescents trying to adjust to a continually changing appearance (Cadogan and Bennum, 2011). There is also a danger for individuals to have unrealistic expectations of surgery, or to believe that aesthetic surgery is the way to solve any psychological or social problems; if such expectations are not met this may evoke psychological distress or the desire for further surgery (Crerand et al., 2013).

Previous research has suggested that during the transition into adulthood, individuals with CL/P may be less successful in relation to education and employment (Ramstad et al., 1995a; Danino et al., 2005), wait longer to get married or to form a long-term relationship (Ramstad et al., 1995a; Danino et al., 2005) and be less likely to have their own children (Yttri et al., 2011). Studies have also suggested that adults with CL/P are at risk of poorer mental health and

lower quality of life than their peers without CL/P (Ramstad et al., 1995b; Marcusson et al., 2001). The genetic component involved in clefting also means there is an increased chance for adults with CL/P to pass the condition onto their children (Mossey et al., 2009).

Interestingly, in spite of, or perhaps because of these challenges, both individuals born with CL/P and their families often report many positive consequences of the condition. Parents have reported a high degree of positive adjustment resulting from their child's condition (Baker et al., 2009). This has included positive perceptions of how they treat others, personal strength, belongingness, affect-regulation, religiousness and (to a lesser extent) optimism and self-understanding (Baker et al., 2009). Further, high levels of social support and approach-oriented coping strategies have been reported (Baker, 2009). For the individual, such consequences have included a high level of satisfaction with appearance (Feragen et al., 2010), particularly in relation to the appearance of physical features unrelated to the cleft (Berger and Dalton, 2009). Individuals born with CL/P have also reported a more positive social environment compared to same-aged peers (Berger and Dalton, 2009; Feragen et al., 2010), along with less emotional distress (Feragen et al., 2010), higher self-esteem and superior quality of life scores (Kramer et al., 2009).

Despite the volume of research in the field of psychological adjustment to CL/P, a number of gaps in understanding are evident and an up to date and authoritative literature is lacking.

Theories and models

In recent years, a collection of theories and models have been presented in an attempt to understand the psychological factors and processes contributing to appearance concerns. The terms 'theory' and 'model' are often used interchangeably; however, theories tend to be broad and more generic in nature, while models are often more specific to a particular aspect (Thompson, 2013). Appearance psychology often draws upon broader theories and models within clinical, health and social psychology, as well as developing and applying models which are specific to appearance, or to a particular condition.

While appearance is rarely the focus of broader theories and models, many can be applied to the field. These include models based on social cognition theory (see Bandura, 1986); self-regulatory theory (see Leventhal et al., 1980); and theories of stress and coping (see Lazarus and Folkman, 1984). When applied to the topic of appearance, models derived from these theories suggest that the manner in which an individual perceives their social world, as well as the way an individual interprets and copes with their condition and any related stressors are important factors in the management of appearance concerns. Examples of ways in which these theories can be applied more specifically to appearance psychology include the discrepancy between an individual's perceived actual and ideal appearance; the relative importance of, and investment in appearance in relation to other personal characteristics; social anxiety arising from the fear of negative evaluation from peers; and attentional and interpretative biases to appearance-related information in the environment (Thompson and Kent, 2001; Cash, 2004; Moss and Carr, 2004; Rosser et al., 2010; Moss et al., 2014).

Models and theories can be useful for developing research plans and interventions, in providing a focus for discussion and debate and in facilitating comparisons between research findings (Rumsey and Harcourt, 2005). However, broader theories and models are often inadequate when trying to understand the complexity of appearance concerns, while appearance- or condition-specific theories and models are often too prescriptive and cannot provide a perspective which is comprehensive enough to fully account for the myriad of variables contributing to individual variation in adjustment. In addition, the success and utility of appearance-specific theories and models is limited until they are more thoroughly tested and refined. Frequently, insufficient attention is paid to the broader social and cultural context when developing theories and models. Although some models allow for change over time or for interaction between different components, the dynamic and fluctuating nature of psychological adjustment remains difficult to capture (Rumsey and Harcourt, 2005). With specific regard to CL/P, appearance is just one component involved in adjustment; the aetiology of the cleft, the perceived burden of treatment, the level of functional impairment and the impact on physical and cognitive development may also play a role. Ultimately, specific theories and models can be useful in guiding research agendas and practice, but are perhaps less useful in terms of understanding the more detailed experience of individuals.

Conceptualising adjustment to CL/P

One way of addressing some of the limitations of individual theories and models is to utilise a conceptual 'framework'. A framework has the ability to take the wider social and cultural context into account and to encompass a wide range of factors, models and theories while also being directly applicable to appearance- and condition-specific concerns. A comprehensive and testable framework, integrating information about the development, maintenance and management of psychological distress for this population would be helpful to both researchers and clinicians and would facilitate investigations and interventions relating to psychological adjustment (Rumsey and Harcourt, 2005). Further, the framework approach allows for the inclusion of findings derived through in-depth qualitative investigation, whereas existing models are wholly informed by quantitative data, further limiting their ability to account for a broad range of experiences.

One such framework was recently developed by the Appearance Research Collaboration (ARC). ARC was formed in 2005 between researchers and clinicians at six different sites across the UK to guide the work of a three-year investigation of the psychosocial factors and processes contributing to successful adjustment to appearance-altering conditions. The factors and processes which were found to play a role in adjustment were described in the form of a conceptual framework, which was designed to inform the development of interventions to promote positive adjustment (see Figure 3). Background factors, such as age, gender, and cultural background can influence the way an individual processes information from the environment. Intervening cognitive processes, such as dispositional style, feelings of social acceptance and appearance valence then play a key role in subsequent outcomes, which may include psychological difficulties such as depression and anxiety, and behavioural outcomes such as aggression and social avoidance. Although this model represents a significant step forward for the field of appearance research and sets a benchmark for future work, further investigation is needed to verify these findings and to validate its utility in clinical practice.

There are a number of existing limitations in the field of CL/P which restrict the opportunity to expand upon the ARC framework, or to develop a similar framework

with a specific focus on this condition. Crucially, there is a distinct lack of consensus in regard to what constitutes a positive psychological outcome for the CL/P population. A wide range of concepts are referred to, and are used inconsistently across studies. Many of the challenges faced by individuals with CL/P throughout life can also be applied to the general population; thus such challenges can be 'normalised' and discussed in relation to the wider context. Unfortunately, CL/P is still heavily medicalised and is widely described as a 'disorder' or an 'anomaly', in which any discrepancies between those with CL/P and their peers without CL/P are pathologised, and in which the cleft is conceptualised as a defining feature of the individual (for example, a 'cleft child', rather than a 'child with a cleft'). Until recently, there has also been a tendency for authors to focus on deficits, differences and risks, rather than strengths, similarities and opportunities. These studies have identified a number of positive consequences of CL/P for both families and the affected individual. Perhaps somewhat surprisingly, these effects have been found to be strongest in those with a visible cleft (CL, CLP) compared to those whose cleft is arguably less visible (CP, SMCP; Feragen et al., 2010). Findings such as these could be indicative of the development of resilience and/or protective factors. Further, authors have begun to note that while individuals with CL/P and their families sometimes report low scores in relation to aspects of adjustment, these are often still within the normal range and/or in line with scores reported by reference groups. So long as deficits and difficulties are the focus of CL/P research, interpretations of findings may be skewed and indicators of resilience may be overlooked. Finally, a lack of exploration of key subgroups of this population limits the potential to develop a holistic and systemic perspective of this field.

Methodological approaches and limitations

In spite of the volume of research published in the area of psychological adjustment to CL/P in recent decades, many gaps in knowledge remain. This is due in part to the inconsistencies reported within the existing literature. While some studies indicate that those born with a cleft experience more psychosocial difficulties than their peers without CL/P, others have found few differences between individuals with CL/P and population-based control groups (see reviews

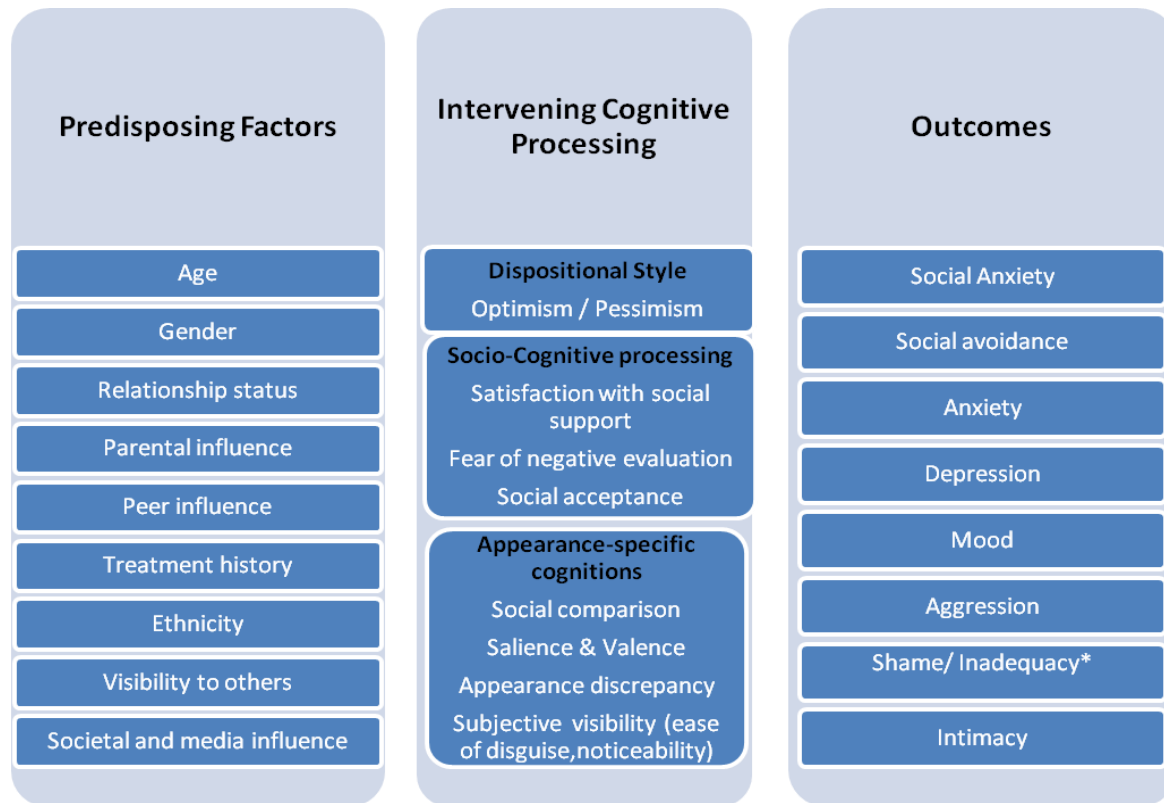


Figure 3: The ARC framework (2009), in Clarke et al., 2013.

such as Turner et al., 1998; Hunt et al., 2005). To further complicate this picture, several studies have found those with cleft to be better adjusted than their peers, and to report a number of positive consequences of growing up with CL/P (Baker et al., 2009; Berger and Dalton, 2009; Feragen et al., 2010; Kramer et al., 2009). While these discrepancies in findings are indicative of the individual variation already described, they also point to a number of methodological challenges.

As is the case with much research, a lack of large enough samples in the field has increased the chance for error and reduced the ability to draw meaningful conclusions. Many studies also lack a control group or appropriate 'norms'. A paucity of longitudinal research in this area means the evidence base is almost entirely reliant upon cross-sectional samples. Ill-defined age groups and the lack of consensus regarding concepts, outcomes and measures have resulted in widespread inconsistency across studies, often rendering useful comparisons between studies unfeasible. Subgroups of patients (for example, those with an identified syndrome, or those from a low socioeconomic group) are often excluded from research in order to meet predetermined biomedical parameters, or neglected altogether. Many authors do not discuss the implications of their

findings, or fail to offer pragmatic suggestions for how the corresponding challenges may be addressed within future research and practice. Finally, there is a relative paucity of literature addressing issues related to CL/P from the perspectives of the affected individual and those around them. In some cases, self-reports are excluded altogether. While both researchers and clinicians have important perspectives to contribute to the knowledge base, a comprehensive understanding of adjustment cannot be achieved without also exploring the patient experience. Recent calls have been made for an increase in qualitative work within the field in order to address this imbalance and to provide insight into the conflicted (and predominantly quantitative) evidence base (Nelson, 2009).

Summary and conclusions

In today's appearance-obsessed world, being different from the 'norm' poses a number of psychological and social challenges. A cleft in the lip and/or the palate is one such visible difference, with the additional potential to impact upon physical functioning, speech development and cognitive development. The burden of engagement with a multidisciplinary care protocol can be considerable and spans from the point of diagnosis into adulthood. In light of these challenges, previous research has been conducted on the psychological impact of CL/P on those affected and their families. However, the current evidence base is conflicted and marred by several conceptual and methodological limitations. While generic and appearance-specific psychological theories and models are useful, they are limited in their ability to incorporate individual variation and to inform the development of appropriate interventions. Conceptual frameworks appear to hold promise, but require further testing and refinement. In order to identify the gaps in the literature, and to consider the impact of some of these conceptual and methodological issues, an authoritative literature review is needed. Additional research to explore some of these 'missing links' may shed further light in relation to the factors and processes involved in psychological adjustment to CL/P.

This doctoral thesis

Aims

The aims of this thesis are to contribute to the understanding of current gaps in knowledge and to offer suggestions for future research and clinical practice through:

- 1) A series of investigations of psychological outcomes of individuals born with CL/P and
- 2) Critical evaluation of current conceptual and methodological approaches in the field and
- 3) Exploration of the experiences and support needs of professionally neglected CL/P patient groups.

Methodology

A variety of methodological approaches and tools were employed, depending upon the research question(s) posed.

Publication 1 discusses the findings of a comprehensive literature review of psychological adjustment in the field of CL/P.

Publications 2 and 3 utilise quantitative statistical methods to investigate differences among patient groups, to compare patient groups to established norms and to test different methodological approaches to analysis.

Publications 4, 5, 6 and 7 employ qualitative interview techniques and inductive thematic analysis to explore new areas of interest in more depth.

Ethical considerations

The research presented in Publication 1 consisted of a literature review and as such did not involve new data collection from human participants.

The research presented in Publications 2 and 3 conformed to guidelines set out by the Regional Committee for Medical Research Ethics, Oslo-East, Norway. Data

presented were based upon a retrospective review and informed consent was provided by the participants' parents.

The research presented in Publications 4, 5, 6 and 7 were reviewed and approved by the Health and Applied Sciences Faculty Research Ethics Committee at the University of the West of England. This research was entirely compliant with the British Psychological Society Code of Ethics and Conduct (2009). All participants were aged over 18 years. Participants provided informed consent and were made aware of their right to withdraw at any time. All data were stored securely and in accordance with the Data Protection Act (1998). Details of appropriate support services were provided.

Structure

Within this doctoral thesis I will demonstrate my ability to meet each of the elements of the university doctoral descriptors via the presentation of one published book chapter, published in a seminal book, Cleft Lip and Palate: Diagnosis and Management, and six peer-reviewed journal articles, published in the leading journal for this field, the Cleft-Palate Craniofacial Journal. Written commentary on each of these publications is provided, and is focused on three key themes: outcomes, methods and neglected groups. Additional evidence to support the wider impact of my work in this field is presented within the appendices.

Presented publications

| Publication no. | Reference |
|-----------------|---|
| 1 | Rumsey, N. and Stock, N.M. (2013) Living with a cleft: Psychological challenges, support and intervention. In: Berkowitz, S., ed. (2013) <i>Cleft Lip and Palate: Diagnosis and Management, 3rd edition</i> . Berlin Heidelberg: Springer-Verlag, pp. 907-915. |
| 2 | Feragen, K.J.B, Stock, N.M. and Rumsey, N. (2014) Towards a reconsideration of inclusion and exclusion criteria in cleft lip and palate: Implications for psychological research. <i>Cleft Palate-Craniofacial Journal</i> . 51(5), pp. 570-579. |
| 3 | Feragen, K.J.B. and Stock, N.M. (2014) When there is more than a cleft: Psychological adjustment when a cleft is associated with an additional condition. <i>Cleft Palate-Craniofacial Journal</i> . 51(1), pp. 5-14. |
| 4 | Stock, N.M. and Rumsey, N. (2015, e-pub 2013) Parenting a child with a cleft: The father's perspective. <i>Cleft Palate-Craniofacial Journal</i> . 52(1), pp. 31-43. |
| 5 | Stock, N.M. , Feragen, K.J.B. and Rumsey, N. (e-pub ahead of print, 2014) "It doesn't all just stop at 18." Psychological adjustment and support needs of adults born with cleft lip/palate. <i>Cleft Palate-Craniofacial Journal</i> , doi: http://dx.doi.org/10.1597/14-178 . |
| 6 | Stock, N.M. and Rumsey, N. (2015, e-pub 2014) Starting a family: The experience of parents with cleft lip/palate. <i>Cleft Palate-Craniofacial Journal</i> , 52(4), pp. 425-436. doi: http://dx.doi.org/10.1597/13-314 . |
| 7 | Stock, N.M. , Feragen, K.J.B. and Rumsey, N. (e-pub ahead of print, 2015) Adults' narratives of growing up with a cleft lip and/or palate: Factors associated with psychological adjustment. <i>Cleft Palate-Craniofacial Journal</i> , doi: http://dx.doi.org/10.1597/14-269 . |

Publication 1

Publication 1

Author commentary

Rumsey, N. and **Stock, N.M.** (2013) Living with a cleft: Psychological challenges, support and intervention. In: Berkowitz, S., ed. (2013) *Cleft Lip and Palate: Diagnosis and Management, 3rd edition*. Berlin Heidelberg: Springer-Verlag, pp. 907-915.

The first step to identifying and addressing some of the conceptual and methodological challenges of the field was an authoritative literature review the area of psychological adjustment to CL/P. Prior to this, the most recent review was conducted in 2005 by Hunt and colleagues. This review identified many inconsistencies within the literature, and highlighted a range of methodological limitations. The primary purpose of Publication 1 was therefore to carry out a comprehensive review of the literature published between 2005 and the present (December 2010), in order to bring this review up to date, to determine whether the field has resolved its methodological problems and to inform future work in this area.

Despite being conducted several years later, the findings of Publication 1 were markedly similar to that of Hunt et al.'s 2005 review. While some studies had reported individuals with CL/P to have poorer psychological and social wellbeing than their peers without a cleft, other studies had found no differences between these groups (see reviews such as Turner et al., 1998; Hunt et al., 2005). In addition, some studies reported individuals with CL/P to have better outcomes than population-based control groups and to report a number of positive consequences of growing up with CL/P (Baker et al., 2009; Berger and Dalton, 2009; Kramer et al., 2009; Feragen et al., 2010). While I believed these findings to be due largely to the complex and fluctuating nature of psychological adjustment, variations in concepts and methodological approaches were likely to play a key role.

Perhaps the most important finding was the number of different concepts and measures being employed. Upon completing the review, I identified multiple competing constructs related to psychological adjustment, none of which were clearly defined, and more than 60 different measures in use. My co-author and I concluded that much of the difficulty stemmed from a lack of agreement as to what constitutes a 'positive psychological outcome'. Without this consensus, the disparate nature of the research and the current inability of clinicians to track patients' progress over time would be unlikely to change.

Prompted by the findings of Publication 1 and in order to tackle the huge number of concepts and measures identified, my co-author and I began working closely with the Cleft and Craniofacial Psychology Special Interest Group (SIG) in 2011. We held a number of discussions around their views of what may constitute a positive psychological outcome, informed by both the literature available to us and the psychologists' clinical experience in the field. Based on this collaborative work and the findings of Publication 1, we then designed a conceptual framework consisting of a short list of broad outcomes, the components of which could then be broken down and measured consistently over time. This allowed us to evaluate the existing measures available in relation to these components and to select the most appropriate tools according to pre-determined criteria, such as psychometric properties, perceived clinical utility and approximate time taken to complete. This framework has since been used to inform the development of a standardised pack of measures for use within research and clinical audit at varying time points through the child's developmental trajectory.

Although this progress was promising, Publication 1 also highlighted a number of additional limitations which had not been discussed to a great degree in previous literature. First, there was a lack of attention being paid to the long-term outcomes of individuals born with CL/P. Research with adults who have grown up with the condition may help to identify the factors and processes which help or hinder psychological adjustment throughout the journey as a whole, and provide a new perspective on the type and timing of interventions. Second, there was an ongoing focus on deficits, differences and points of risk. It was clear that the patient perspective was largely missing, as was the insight of how to facilitate the challenges of living with a medical condition into a strengthening experience for all involved. Third, individuals with CL/P had often been treated as a homogeneous

group, with little consideration of possible confounding variables and potentially vulnerable subgroups. In addition, there was a heavy bias towards the experiences of mothers of children born with a cleft, with few attempts to involve fathers in research; a lack of understanding of the impact of additional conditions related to CL/P (such as developmental delay and AD/HD); and few attempts to address the potentially unmet needs of adults with CL/P no longer receiving routine treatment.

This work was presented at the annual conference of the Craniofacial Society for Great Britain and Ireland (CFSGBI) in 2011 and was awarded the President's Medal for contribution to the field. The work was also presented at the 9th European Craniofacial Congress in Salzburg, Austria in 2011. The review was updated prior to publication in the seminal book "Cleft Lip and Palate: Diagnosis and Management (3rd edition)" edited by Dr Samuel Berkowitz in 2013.

Publication 1 is presented within this thesis as an introduction to the existing state of the literature, and as an identification of the significant gaps in current knowledge. It draws into focus the primary aim of this DPhil; to investigate why such gaps exist and to address some of these methodological and conceptual limitations, while providing clear suggestions for future research and clinical practice.

Publication 2

Publication 2

Author commentary

Feragen, K.J.B., **Stock, N.M.** and Rumsey, N. (2014) Toward a reconsideration of inclusion and exclusion criteria in cleft lip and palate: Implications for psychological research. *Cleft Palate-Craniofacial Journal*. 51(5), pp. 570-579.

I met Dr Feragen, Clinical Psychologist for the Cleft Lip and Palate Team in Oslo, Norway, at the CFSGBI conference in 2011, where I presented the findings of Publication 1. Shortly after this, Dr Feragen was awarded a three-year research grant which also gave her the opportunity to travel to the UK to work at CAR for one year. During this time, Dr Feragen asked me to collaborate with her on a paper, which would attempt to address some of the key conceptual and methodological challenges I had highlighted in Publication 1. Over eleven consecutive years, Dr Feragen had collected data on psychological adjustment from children and their parents visiting the cleft clinics in Oslo. This accumulated data would not only give us one of the largest CL/P samples to date, but would allow us to look at important subgroups of children who would normally be excluded from research.

Within previous research, children with a known syndrome or severe developmental/neurological difficulties have been excluded from CL/P studies, in an attempt to ensure homogeneity within samples. Unfortunately, the biomedical parameters which are used to identify and exclude these children are rarely implemented in a uniform way across studies. Furthermore, additional conditions such as autism, attention deficit/hyperactivity disorder (AD/HD), specific language impairment, dyslexia, learning difficulties and global developmental delay are known to impact on psychological development in the wider literature, yet children with these conditions are included in CL/P samples.

Publication 2 employed quantitative analysis to investigate the effect of two different analytical approaches on the same data. The first analytical approach used conventional exclusion criteria, excluding those children with a known syndrome or severe developmental/neurological diagnosis. The second analytical

approach included all children irrespective of diagnosis, while also taking into account the presence or absence of a variety of additional conditions. Results between the two analytical approaches were compared using the psychological outcome measures employed in the Oslo cleft clinic. The statistical analysis produced a large set of interesting results; however, Dr Feragen and I were concerned that we might lose focus and clarity by addressing multiple research questions in one paper. After discussing our options with third author Professor Rumsey, we decided to divide our findings across two papers; one would focus on methodology, while the second would concentrate on the subgroup(s) identified.

The findings of Publication 2 suggested that different outcomes regarding the psychological adjustment of children with CL/P are evident depending upon the type of analytical approach used. Specifically, when using the traditional biomedical approach to exclusion, results were indicative of a high degree of psychological difficulties in children with CL/P. When using the suggested alternative approach, results suggested that children with a cleft alone reported scores which were in line with the reference group, while a high degree of psychological difficulties were found in children with CL/P and one or more additional conditions. It is therefore possible that the combination of poorly-defined and inconsistently applied exclusion criteria and the failure to identify those children with a range of additional conditions may be affecting the findings produced and the conclusions drawn in previous studies. Ultimately, this is likely to contribute to the conflicting results evident within current literature.

Publication 2 emphasises the importance of careful assessment and reporting of all conditions which are present in addition to the cleft, in order to distinguish between the psychological impact of CL/P and the psychological impact of additional conditions. This alternative approach throws into question the previous reliance upon using biomedical parameters for psychological research. In addition, the use of control groups, cut-off scores and/or norms is essential, to allow for discussion of the ways in which children with CL/P compare to their peers, rather than the simple reporting of scores without an appropriate reference. Without these comparisons being possible in Publication 2, findings would have been more difficult to interpret and thus more difficult to apply in practice.

The implications drawn from Publication 2 were presented at the CFSGBI conference in 2013 and published in the leading academic journal for this field, the Cleft Palate-Craniofacial Journal, in 2014.

Although Publication 2 provided a clear example of how the conceptual and methodological limitations may currently thwart the field, as well as offering suggestions for an alternative approach, the division of the findings into two separate research questions meant that an in-depth look at the subgroup(s) which would normally be excluded was still missing. Publication 3 aimed to explore these groups in more detail.

Publication 3

Publication 3

Author commentary

Feragen, K.J.B. and **Stock, N.M.** (2014) When there is more than a cleft: Psychological adjustment when a cleft is associated with an additional condition. *Cleft Palate-Craniofacial Journal*. 51(1), pp. 5-14.

Several studies in this area have reported a relatively high frequency of additional conditions, such as those described in Publication 2, among children with CL/P. Despite this, virtually no studies have investigated the psychological impact of these conditions on individuals with CL/P, nor discussed how these conditions should be approached clinically. To follow on from the first collaborative publication between Dr Feragen and I, this paper utilised quantitative methods to investigate the impact of having an additional condition on psychological adjustment in children with CL/P at age 10.

As expected, a high percentage (39.5%) of the children in our sample had at least one condition in addition to the cleft. These children reported significantly greater psychological difficulties in relation to social experiences, attention, emotional problems and behavioural conduct than their peers with a cleft alone and in comparison to the reference group without CL/P. In stark contrast to some of the findings detailed in previous literature, children with a cleft alone reported adjustment scores which were within the normal range and that were similar to those reported by the reference group.

One essential finding of both Publications 2 and 3 is that in spite of the psychological challenges associated with CL/P, children with a cleft alone reported scores in line with the reference group. First, this implies that the cleft alone may not constitute a significant risk factor, at least at age 10, and throws into question the tendency for the cleft itself to be conceptualised as a definitive pathology. Thus, potential confounding factors which may increase the risk of psychological distress in those with CL/P should be carefully considered, instead of assuming outcomes are homogenous and apply to the group as a whole. Another interpretation is that protective factors play a key role in the psychological

adjustment of children with CL/P. Of equal importance, the findings highlight that those children with an additional condition may represent a high risk subgroup. Early identification of children who exhibit signs of developmental difficulties, as well as the integration of appropriate support for these families, may be crucial for psychological adjustment among this vulnerable subgroup.

Publication 3 also highlighted a number of methodological implications. For example, the decision was taken to look at one age group only when addressing the research question. Although this approach meant that fluctuations in the relative impact of an additional condition and the psychological adjustment of those with CL/P alone could not be addressed in Publication 3, it also removed the potential for age to confound results, a common limitation evident in previous research. Second, even with a relatively large overall sample the total number of children included in the analyses is reduced when looking at subgroups. Publication 3 therefore demonstrates the importance of large and representative samples in order to facilitate the investigation of subgroups within CL/P samples. Finally, differences between self-reports and parent-reports were observed. Therefore, both self- and parent-reports may be needed to conduct a comprehensive assessment of the child's (and the parents') psychological adjustment, rather than rely on one or the other as has been done previously.

The findings of Publication 3 were presented at the CFSGBI conference in 2013. The paper was accepted for publication in the *Cleft Palate-Craniofacial Journal* on its first review just one month after submission, and was published in 2014.

Publication 3 implied that a lack of exploration of important subgroups may have contributed to the conflicting findings within the current literature. It also provided some additional methodological insights, such as the need to include a range of different perspectives when investigating psychological adjustment. One of the strengths of Publication 3 was the inclusion of measures completed by both the mother and the father. To date, little research has included fathers in research, or looked explicitly at fathers' experiences of CL/P and potential support needs. Fathers of children with CL/P thus represent another unexplored subgroup in the field which may provide a different perspective on the psychological adjustment of individuals with CL/P and their families.

Publication 4

Publication 4

Author commentary

Stock, N.M. and Rumsey, N. (2015, e-pub 2013) Parenting a child with a cleft: The father's perspective. *Cleft Palate-Craniofacial Journal*. 52(1), pp. 31-43.

While conducting the literature review in 2010, I identified a paucity of research with fathers of children born with CL/P. I found this surprising, mainly due to the volume of literature discussing the impact of CL/P on 'parents', which then went on to discuss the mothers' views only. While many of these authors had not considered the possible limitations of excluding fathers from their investigations, others had commented that fathers were 'difficult to recruit into psychological studies'.

From studying research on parent-child relationships, I found that the role of the father is different to that of the mother (for a review see Lamb, 2010). Specifically, fathers spend a much higher percentage of their interaction with their child engaging in stimulating, playful activity than do mothers. From these interactions, children learn to how to regulate their emotions and behaviour. Fathers also tend to promote independence and orientation to the outside world, which in turn is likely to exhibit self-control, achievement and pro-social behaviour in the child. An absent or incapacitated father therefore impacts significantly on the child's physical and emotional developmental trajectory and influences the wellbeing of the family unit as a whole. Furthermore, the role of the father cannot be replaced by another family member. Following a literature review by Nelson and colleagues in 2012, which called for the implementation of improved psychological support for families affected by CL/P, I decided to explore the experience of having a child with CL/P from the perspective of the father. Until this perspective had been heard, any familial support was likely to be limited in its scope.

In order to adopt the inclusive approach advocated by the findings of Publication 2, I chose not to exclude any prospective participant on the basis of age, cleft type, ethnicity, additional conditions or other background variable. I also decided to conduct the interviews over the phone, to eliminate travel time and cost, and to

allow for flexibility regarding where and when the interview could take place. I hoped this would help to overcome the difficulty of recruiting fathers into the study, as stated by previous research, and would also allow me to recruit fathers from across the UK, without any geographical limitations.

In terms of my own development, this was one of the first pieces of research I had instigated, designed, conducted, analysed and disseminated with minimal supervision. I thoroughly enjoyed the process, which also enlightened me to the potential power and significance of qualitative approaches. For example, rather than dictate what I wanted to measure, based on what I guessed to be important, I took an inductive approach to interviewing and allowed the participants to simply tell their story in their way, with very occasional prompting and follow-up from me. Contrary to the previous and widespread claim that men are difficult to recruit into psychological research studies, I had recruited and interviewed fifteen fathers in under two months, and collected a wealth of informative data. This, along with the power and emotion with which each story was told, made me feel sure I had tapped into something important.

The resulting data suggested that while fathers' reported experiences are comparable to those previously described by mothers, the support that is available for fathers, both in relation to having a baby with CL/P and to pregnancy in general, is considerably less. Fathers described their primary role to be to support their partner and family both practically and emotionally. Unfortunately, without an outlet for their own concerns and emotions, this seemed to put a great deal of strain on their own health. The findings of Publication 4 thus point to the importance of offering psychological support to fathers and of tailoring this support to fathers' specific practical and emotional needs, as well as adopting a more inclusive approach to care more generally. Incorporating fathers' views into future research would provide an additional and important perspective on psychological adjustment to CL/P, with the potential to shed light on conflicting reports in the literature. Further, this approach would promote a more holistic and systemic view of adjustment.

The findings of Publication 4 also point to a significant variability in need, both between participants and at different stages of the treatment pathway. In addition, fathers commented that although contact with the cleft team seemed to trail off after the initial surgeries had been completed, the need for support could arise at

any time. This highlights the importance of a stepped approach to care (see Publication 1) in which different levels of, and approaches to, intervention are made available on a flexible basis throughout the family's entire journey. This finding is also an example of how individual differences and fluctuations in adjustment over time could have affected the findings published in previous literature. One limitation of the paper was the heterogeneity among the recruited sample. While this matters less in terms of the quality of qualitative data compared to quantitative data, it was clear that a larger sample (given fewer time restraints) would have been beneficial. Nonetheless, Publication 4 demonstrated that men (and potentially other previously neglected groups) can be recruited into psychological research, if the approach and the research question are appropriately formulated.

The findings of Publication 4 were presented at the CFSGBI conference in 2012 and at the 12th International Craniofacial Congress in Orlando, USA in 2013. The results were also disseminated directly to participants, as well as to all members of the Cleft Lip and Palate Association (including parents and health professionals). I co-wrote a UWE national press release which received considerable online interest and some local media attention, including a fifteen-minute discussion on BBC Radio Bristol during the prime listening hour. The final paper was published in the Cleft Palate-Craniofacial Journal in 2015 (e-pub 2013). The findings have since been picked up and disseminated by additional charities in the UK, Australia and New Zealand.

Publication 4 highlighted the value of the patient perspective in informing research agendas and clinical practice and demonstrated the potential to recruit 'hard to reach' and geographically dispersed participants. Another 'hard to reach' and professionally neglected subgroup of patients was adults with CL/P. Again, the patient perspective appeared to be lacking, and it seemed as though adults with CL/P could be experiencing psychological challenges that had not yet been fully recognised or provided for by cleft services. Publication 5 thus aimed to explore this subgroup in more depth.

Publication 5

Publication 5

Author commentary

Stock, N.M., Feragen, K.J.B. and Rumsey, N. (e-pub ahead of print, 2014) "It doesn't all just stop at 18." Psychological adjustment and support needs of adults born with cleft lip/palate. *Cleft Palate-Craniofacial Journal*, doi: <http://dx.doi.org/10.1597/14-178>

Having thoroughly enjoyed the qualitative process of Publication 4 and found it extremely useful for eliciting rich data in areas where little is known, I decided to approach another gap in the knowledge in a similar way. The initial review (Publication 1) had identified a significant lack of literature in relation to long-term outcomes for individuals with CL/P, and this finding seemed concurrent with the tendency for routine cleft treatment to conclude around the age of 18 years. When critically evaluating the studies which had been published in relation to adults with CL/P, the existing literature was overwhelmingly negative, largely out of date and overly focused on objective medical outcomes. The patient perspective was distinctly lacking; through my work with CLAPA I had met many adults with CL/P and felt there was another side to the story.

As before, I chose not to exclude anyone on the basis of background variables, to ensure I gathered as many different perspectives as possible. Learning from the participant heterogeneity found in Publication 4, I decided to recruit a larger sample this time, in an attempt to collect data from a more representative group of adults. However, I was also aware that recruiting participants who were no longer receiving cleft treatment and who were geographically dispersed would be a challenge. I therefore employed a number of different and creative recruitment strategies, including a university press release which was picked up widely by local, national and online media, advertisements through CLAPA and related organisations, and direct email advertisements to those eligible adults who had previously enquired about other cleft research. As soon as the adverts were released, I was inundated with messages from adults who wanted to take part in the study. Again, I felt this was testimony to the fact that this was a subject which warranted in-depth investigation. In just six weeks, I had interviewed 52 adults with CL/P. Despite having more adults contacting me every day to take part, I

chose to stop interviewing at this point and gather my thoughts on the data I had. The result, unsurprisingly, was an almost insurmountable quantity of qualitative data. While the data was incredibly valuable, I began to feel as if I was drowning in it, and I struggled to comprehend where I might go next. Driven by a desire to do justice to the stories I'd been told, I eventually decided to divide the data into three papers (Publications 5, 6 and 7). Since qualitative analysis of the data resulting from the interviews had proved difficult, and in order to further involve 'patients' in the research process, I chose to collect feedback on the preliminary findings. I hosted a presentation and discussion at a CLAPA workshop for people affected by CL/P and their families, as well as to the CLAPA Adult Voices Council in 2013. I also sent a summary to, and collected feedback from the participants themselves.

The resulting publication detailed five key themes which adults with CL/P identified as critical to their wellbeing in adulthood from their perspective. It explored issues in relation to further treatment, social and romantic relationships, higher education and employment and access to psychological support. Contrary to many of the previous studies carried out with this population, Publication 5 suggested that the majority of adults with CL/P in this study had adjusted well to the challenges associated with their condition and reported many positive outcomes. This may in part explain the discrepancies within the existing literature and emphasises the importance of including the patient perspective in research and practice. Nonetheless, Publication 5 implies that some issues attributed to CL/P may continue into adulthood, and new issues arising later in life, such as employment, long-term relationships and starting a family, may warrant further investigation and additional psychological support. Publication 5 discussed the experiences and identified the felt needs of a previously neglected group who have rarely had their voice heard. The findings have clinical implications for adults who have already left the cleft service, as well as the children who are current engaged in the treatment pathway.

In 2014, the final results of Publication 5 were disseminated to the CLAPA membership, as well as to the wider community through the use of online media. Based on the findings of this research and following a formal collaborative proposal written by myself and the CLAPA Adult Voices Council, CLAPA agreed to select 'adults with CL/P' as the theme for the 2014 Cleft Awareness Week, and

invited me to be part of the campaign committee. Later in the year, Publication 5 was presented at the biannual Appearance Matters conference in Bristol, UK in 2014, in collaboration with the Co-Chairs of the CLAPA Adult Voices Council, one of whom also participated in the original study. The study thus provided an example of how 'Patient and Public Involvement' (PPI) in research can help to develop and improve the quality of research and dissemination. The final paper was published online in the Cleft Palate-Craniofacial Journal (e-pub ahead of print) in 2014. The findings have since been picked up and disseminated by charities in the UK, Australia and New Zealand. The printed version is due to appear in the September 2015 issue of the journal and has been selected for a National press release (USA) and awarded open-access status.

As a result of dividing the data into three papers, Publication 5 was unable to detail adults' experiences and views in relation to starting their own family, a potentially difficult and largely unexplored area, due to the genetic component involved in CL/P. Publication 6 thus aimed to describe this experience from the patient perspective.

Publication 6

Publication 6

Author commentary

Stock, N.M. and Rumsey, N. (2015, e-pub 2014) Starting a family: The experience of parents with cleft lip/palate. *Cleft Palate Craniofacial Journal*, 52(4), pp. 425-436. doi: <http://dx.doi.org/10.1597/13-314>.

While working on Publication 4, I interviewed one father who had been born with CL/P himself. The possibility of future generations being affected by CL/P was also raised in Publication 5. Due to the genetic component involved in clefting, there is an increased chance of an individual's children (and subsequent generations) also being affected (Mossey et al., 2009), although the genetic and environmental mechanisms involved are poorly understood.

As part of the larger adults study, I had already decided to advertise specifically for adults who were now also parents, in order to explore how they had felt about starting their own family. I recruited adults who had children born either with or without CL/P, which would allow me to see if there were any differences between these groups of parents, which had not been done before. I chose not to interview parents who did not have a cleft themselves, since there was a wealth of literature already available on this.

While searching the existing literature, I found two studies that had attempted to compare the psychological adjustment of groups of parents with and without their own diagnosis of CL/P (Andrews-Casal et al., 1998; O'Hanlon et al., 2012). Although both studies concluded that psychological support was necessary for parents who had CL/P themselves, both studies failed to find significant differences between parents with and without CL/P on a number of quantitative measures. Additionally, neither study had provided clear suggestions of the type of support that may be required. However, the study by O'Hanlon and colleagues (2012) had included a qualitative component, which suggested that parents with and without their own diagnosis of CL/P may experience and react to their child's diagnosis in different ways, and seemed to add insight to the lack of findings within the quantitative component of the study. I therefore felt this topic warranted further

investigation, in order to explore the potentially unmet needs of this unique group from their own perspective, to more fully understand some of the reported discrepancies in the literature and to provide suggestions of how to support this subgroup of patients in practice.

The findings of Publication 6 suggested that although a parental diagnosis of CL/P may be felt by interviewees to impact parents' experiences of having a child, these parents do not necessarily experience elevated levels of distress as a result. In fact, incorporating their own experiences of growing up with a cleft into their style of parenting may be considered advantageous for both the child and the parent. Publication 6 therefore advocates a move away from 'differences' and 'comparisons' (as investigated in the two previous studies on this topic) to a more holistic and contextual standpoint. Publication 6 also demonstrates how CL/P can be a mechanism for growth and bring an individual's strengths to the fore. The findings suggest that an ongoing cycle of psychological adjustment may be developed and maintained over generations. Finally, Publication 6 strongly highlights the importance of information for prospective parents regarding the heritability of CL/P, and of the need for access to genetic counselling and psychological support if required.

Publication 6 was presented at the CFSGBI conference in 2014, and disseminated to the CLAPA membership and via online media. The findings of the research were also included in the 2014 Cleft Awareness Week campaign. The final paper was accepted after the first review and published online in the Cleft Palate-Craniofacial Journal in 2014. The findings have since been picked up and disseminated by charities in the UK, Australia and New Zealand. The article appeared in print in July 2015 and was selected for a National press release (USA) and awarded 'open access' status. A number of UK cleft teams have since revised their 20-year audit clinic to include information about heritability for prospective parents with CL/P.

While Publications 5 and 6 provided an in-depth and patient-centred perspective on issues specific to adulthood, a look at the patient journey as a whole was still lacking. Publication 7 aimed to identify factors and processes which may contribute to psychological adjustment according to the patient perspective.

Publication 7

Publication 7

Author commentary

Stock, N.M., Feragen, K.J.B. and Rumsey, N. (e-pub ahead of print, 2015) Adults' narratives of growing up with a cleft lip and/or palate: Factors associated with psychological adjustment. *Cleft Palate-Craniofacial Journal*, doi: <http://dx.doi.org/10.1597/14-269>.

When interviewing adults about their experiences of CL/P, I not only asked them about issues in adulthood, but also what they could remember from growing up with the condition. After much consideration, I had decided not to confuse Publication 5 with reported experiences of childhood and adolescence, due to the volume of data accumulated and the potential for multiple research questions. However, I felt this story provided an interesting and thus far absent perspective and still needed to be told in a separate paper.

Initially, my aim for the paper was to use this remaining data to inform the development and evaluation of new and existing interventions in the field. One of the difficulties of psychological intervention is that there is little clear guidance about what the interventions should target, and even fewer studies which provide evidence for the worth of existing interventions (Jenkinson et al., under review; Norman et al., in press 2014; Bessell and Moss, 2007). A recent priority setting initiative by the James Lind Alliance (www.lindalliance.org) identified the optimal type and timing of psychological intervention to be the most important unanswered research question in CL/P. I believed the data I had collected could provide a starting point by identifying some of the key factors which may contribute to the development of psychological distress and resilience across the lifespan.

Using the ARC framework of adjustment as inspiration (see Figure 2), I began to thematically analyse the remaining data on adults according to factors which had seemed to help or hinder psychological adjustment. I identified three main themes: background factors (characteristics that are relatively constant); external factors (aspects that are largely influenced by other people or by the environment); and internal psychological factors (perceptions, interpretations and attributions

made by the individual). While some of these factors were contextual, many others appeared to be amenable to change, and thus, to psychological intervention. The paper went on to discuss these findings in the context of a broad review of the relevant literature, pertaining to adjustment in CL/P, visible difference and health psychology more generally.

I felt confident in and proud of this paper and its potential, so I was surprised to receive a mixed review from the Cleft Palate-Craniofacial Journal shortly after submission. While one reviewer was very positive about the paper, the second reviewer had significant concerns about the scope and aims of the paper as a whole. Given the discrepancies between the two reviews, it was difficult to respond, and I was a little disappointed not to receive additional guidance from the editor. Gradually though, I began to realise that the scope of the paper was possibly too ambitious. Although the paper did have the potential to inform interventions, this related more to a possible future outcome than an initial aim. Additionally, the paper could only provide the limited perspective of the individuals in the recruited sample. It is known from the general psychology literature (see Cantor and Kihlstrom, 1987) that individuals try to find meaning in their experiences and that perceptions of events can fluctuate over time. Thus, patient theories regarding their own development are not necessarily psychologically accurate or the basis for interventions. However, after some reformulation I was able to express that the strength of the paper was to provide a patient perspective, which could potentially be added to the knowledge derived from previous research and the clinical experience of the psychologists working in CL/P. This in turn could provide collective insight into the factors and processes which may contribute to adjustment in the CL/P population. Additionally, the scope of the discussion section of the paper was significantly reduced and refined, in order to bring more focus to the paper and to ensure the findings were not over-generalised.

The final version of the paper described and discussed each of the patient-identified factors and the possible implications for future research and practice. Above all, the findings illustrated the potential degree of individual variation in patient perspectives, identifying positive, neutral and negative responses from participants in some cases. This finding underlined the importance of psychological variables and cognitive processes in adjustment to CL/P and may explain a proportion of the conflicting results reported in the existing literature. In

addition, several issues which are also relevant to the general population were identified, such as age, gender, societal pressures, optimism and faith/spirituality. These findings highlight the difficulties in trying to tease apart those challenges pertaining to the general population and those challenges specific to, or exacerbated by, CL/P and its treatment, as well as understanding how these challenges may interact. Publication 7 also demonstrated the potential to learn from those patients who report having adjusted well to the challenges associated with CL/P, in addition to those who present with difficulties attributed to CL/P. The number and breadth of psychological factors identified in this study is testament to the importance of psychology in the field of CL/P and demonstrates a clear role for psychologists in tackling appearance-related concerns, designing information and self-help materials, supporting patient decision making and exploring ways of improving social interaction, as well as providing specialist psychological support.

The findings derived from Publication 7 were presented at the CFSGBI conference in 2015 and published in the *Cleft Palate-Craniofacial Journal* (e-pub, 2015). The findings have since been picked up and disseminated by charities in the UK, Australia and New Zealand.

Future challenges for the field of CL/P include deciding how and when to measure the factors contributing to adjustment, and ensuring that this approach is carried out consistently across different teams and organisations where possible. Longitudinal research which is able to identify the optimal type and timing of psychological interventions for individuals with CL/P and their families, and the involvement of clinician and patient representatives in research, is essential to this effort.

Synthesis and conclusions

Summary of themes

This doctoral thesis began by presenting an overview of the current state of knowledge in the field of psychological adjustment to CL/P and identified a number of significant gaps in the evidence base (Publication 1). The publications that followed (Publications 2-7) attempted to address some of these gaps in relation to three key themes: psychological outcomes, research methods and professionally neglected groups.

Outcomes

In the field of visible difference, findings indicate that adjustment to an appearance-altering condition is multifaceted, involving a complex interplay of physical, cultural, psychological and social factors (Moss, 1997; Clarke 1999; Endriga & Kapp-Simon, 1999; Thompson & Kent, 2001; Rumsey & Harcourt, 2004). Although this approach is now widely accepted, a clear lack of consensus in regard to what constitutes a positive psychosocial outcome, specifically within the field of CL/P, and how to measure this outcome, still exists. This lack of direction was clearly highlighted in Publication 1 and suggested a framework approach as a first step toward achieving consensus. The findings of this literature review instigated an ongoing working relationship with the Cleft and Craniofacial Psychology SIG, from which a conceptual framework and a corresponding set of clinically useful and psychometrically robust measures was produced (see Figure 4 and Table 1 below).

A number of previous studies have identified a high prevalence of cognitive difficulties (such as autism, AD/HD, dyslexia, specific language impairments and developmental delay) among individuals with CL/P (Richman et al., 2012; Roberts et al., 2012). The findings of Publications 2 and 3 confirmed this prevalence within a large CL/P sample and strongly suggested that these additional difficulties could be expected to impact upon psychological outcomes. Specifically, children with a condition in addition to their cleft report significantly worse psychological outcomes at age 10 in comparison to both their peers without a cleft and those children with

a cleft alone. This clearly implies the need for early identification and subsequent support for this potentially vulnerable subgroup of children and their parents, and suggests that the cleft itself may not constitute a significant risk factor, at least at age 10.

Previous research focussing on the long-term outcomes for individuals born with CL/P has highlighted a range of deficits and difficulties in comparison to unaffected peers (Ramstad et al., 1995a; Ramstad et al., 1995b; Marcusson et al., 2001; Danino et al., 2005; Yttri et al., 2011). To address psychological adjustment to CL/P in adulthood from an alternative view, Publications 5 and 6 investigated the long-term outcomes for individuals born with CL/P from a patient-led, qualitative perspective. These papers identified adults' perceived needs for treatment information, guidance and psychological support beyond the age of 18 years, with particular emphasis on certain life events, including higher education and employment, the development of intimate relationships and the potential impact of CL/P on future generations. Equally, the studies demonstrated the potential for positive psychological adjustment to be developed and maintained, in spite of any challenges.

Previous reviews of the appearance field have concluded that the degree of individual variation within adjustment appears to be high, but remains poorly understood (Rumsey and Harcourt, 2005). In response to this, Publication 7 identified and discussed numerous factors and processes which may contribute to psychological adjustment according to the differing perspectives of individuals with CL/P. The findings provide a unique and valuable point of view of the degree of variation between individuals and this could be measured in the future. In addition, the findings offer insight into how CL/P may be turned into a strengthening experience for all involved, and how optimal outcomes may be achieved.

Methods

Within the existing literature to date, findings are conflicting and conclusions are difficult to draw. While some studies indicate that those born with CL/P experience more psychological difficulties than their peers without a cleft, others have found

| Predisposing factors | Early indicators (0-3 years) | Key domains during childhood (4-11 years), adolescence (12-17 years) and adulthood (18+ years) |
|---|--|---|
| <p>Genetic (including a family history of CL/P and the presence of additional conditions and/or syndromes)</p> | <p>Healthcare (including perceived burden of treatment, satisfaction with care received and identification of co-morbid conditions)</p> | <p>Social functioning (including perceived teasing/bullying and social anxiety)</p> |
| <p>Demographic (including gender, ethnicity and socioeconomic status)</p> | <p>Parental wellbeing (including levels of stress, anxiety and depression and appraisals of CL/P)</p> | <p>World view (including dispositional style, perceptions of stigma and locus of control)</p> |
| <p>Familial factors (including medical history, family functioning and cultural/religious background)</p> | <p>Parent-infant interactions (including child temperament and quality of attachment)</p> | <p>Appearance (including subjective satisfaction with appearance, salience and valence)</p> |
| | | <p>Vocational milestones (including cognitive development, educational experience and satisfaction with employment)</p> |
| | | <p>Psychological wellbeing (including perceived quality of life and self-esteem)</p> |
| | | <p>Condition-specific factors (including experience and expectations of treatment, perceived impact of CL/P on life and appreciation of positive growth)</p> |

Figure 4: A conceptual framework of psychological adjustment to cleft lip and/or palate.

Table 1: Consensus achieved to date: outcome measures now being used in The Cleft Collective Cohort Studies and the National CL/P audit at age 5 years.

| Measure | Related domains | Generic or specific | Parent or child-based | Time points |
|--|---|----------------------------|------------------------------|---|
| Life Events Scale | Parental wellbeing | Generic | Parent | Diagnosis/birth 18 months 3 years 5 years 8 years |
| Pediatric Quality of Life - Family Impact module (PedsQL FI) | Familial factors Parental wellbeing | Generic | Parent | Diagnosis/birth 18 months 3 years 5 years 8 years |
| Pediatric Quality of Life - Healthcare Satisfaction module (PedsQL HS) | Healthcare | Generic | Parent | Diagnosis/birth 18 months 3 years 5 years 8 years |
| Revised Life Orientation Scale (LOT-R) | Parental wellbeing World view | Generic | Parent | Diagnosis/birth 3 years 5 years |
| Perceived Stress Scale (PSS-10) | Parental wellbeing | Generic | Parent | Diagnosis/birth 18 months 3 years 5 years 8 years |
| Hospital Anxiety and Depression Scale (HADS) | Parental wellbeing | Generic | Parent | Diagnosis/birth 18 months 3 years 5 years 8 years |
| Your Child's Cleft* | Condition-specific factors | Cleft-specific | Parent Child | Diagnosis/birth 18 months 3 years 5 years 8 years |
| Ages and Stages (cognitive development) (ASQ-3) | Vocational milestones | Generic | Child | Diagnosis/birth 18 months 3 years 5 years |
| Ages and Stages (socio-emotional development) (ASQ-SE) | Social functioning Psychological wellbeing Parent-infant interactions | Generic | Child | Diagnosis/birth 18 months 3 years 5 years |
| Strengths and Difficulties Questionnaire (SDQ) | Psychological wellbeing Vocational milestones Social functioning | Generic | Child | 5 years 8 years |

those with CL/P to be better adjusted than their unaffected peers, or have reported few differences (see reviews by Turner et al., 1998 and Hunt et al., 2005). To explore further why this may be the case, Publication 1 updated these previous literature reviews while highlighting some of the key methodological challenges and limitations in this field. This included the dependency upon small and heterogeneous samples followed by the tendency to treat such samples as if they were homogeneous, and an over-reliance on 'objective' measurement coupled with a comparative lack of research exploring the patient perspective.

Traditionally, and in line with a biomedical approach to research, subgroups of patients (including those with a genetic syndrome) are normally excluded from CL/P samples. However, these criteria are not often applied consistently or well described. Additionally, children with associated conditions remain included in the sample, despite the potential for the condition to affect psychological adjustment and therefore skew results. Publication 2 draws attention to the idea that the type of, and approach to inclusion/exclusion criteria in scientific research may distort the findings that are presented and the conclusions that are drawn. Using psychological adjustment in CL/P as an example, the paper demonstrated that when all children with CL/P are included and are categorised according to diagnoses, children with a cleft alone report scores which are within the normal range, and are similar to those of their peers without CL/P, while those with a condition in addition to the cleft report higher levels of psychological difficulties. Thus, there is a need to accurately and consistently assess and document the presence of an additional condition (as well as other potentially confounding variables) to allow researchers to explore the data according to relevant subgroups and to provide a better theoretical account of these interactions. In order to fully achieve this, large samples are needed. In addition, the use of control groups, cut-off scores and/or norms is essential, to allow for discussion of the ways in which individuals with CL/P compare to their peers, rather than the simple reporting of scores without an appropriate reference group. Publications 2 and 3 also identified discrepancies between self- and parent-reports of psychological functioning. Discrepancies between parents, professionals, teachers and patients have been highlighted in previous literature (Goodman, 2001; Foo et al., 2013; Alansari et al., 2014), and call for the inclusion of multiple perspectives in both research and practice. These findings also highlight the need to restrict the age range of participants in a given sample.

Publications 4, 5, 6 and 7 validate the use of patient-focused qualitative approaches in this (and other) medical field(s), as originally emphasised by Nelson (2009). Where little is known about a subject, or if exploring a topic from a new perspective, qualitative data can provide insight and form a basis for future quantitative work. Additionally, when quantitative studies produce conflicting findings, qualitative approaches may help to interpret these discrepancies. Publications 4, 5, 6 and 7 also highlighted the value of Patient and Public Involvement (PPI) in research in improving the quality of the research and its subsequent dissemination (see INVOLVE for more information: www.invo.org.uk).

Neglected groups

This thesis has argued that, while subgroups of patients and families continue to be excluded or neglected in CL/P research and practice, our understanding of adjustment to CL/P and how to optimise outcomes will remain incomplete.

The review outlined in Publication 1 identified a number of important patient groups who have traditionally been excluded from research for methodological reasons, or who have largely been professionally neglected altogether. These patient groups included: children with additional conditions (Feragen, 2010), fathers of children with CL/P (Nelson et al., 2012), adults with CL/P (Marcusson et al., 2001) and families with a history of CL/P (Andrews-Casal et al., 1998; O'Hanlon et al., 2012).

The collection of work presented in this DPhil thesis (Publications 2-7) has made the first step toward offering suggestions for incorporating these subgroups into future research and in caring for them in practice, in support of a more holistic and systemic approach.

Applicability of the findings to psychological theory

Frameworks

The process of building a framework for CL/P has verified the outcomes which we believe to be important in psychological adjustment to CL/P, and has provided the opportunity to track the progress of related variables across an individual's lifespan

(see Figure 4 and Table 1 above). The process has also provided support for the suggestion that individual models and theories cannot do justice to the complexity of individual experience, and has confirmed the value of utilising an overarching 'framework' approach. The work presented in this DPhil has shown that a framework approach has the ability to take the sociocultural context and relevant qualitative findings into account and to encompass a wide range of factors, models and theories while also being directly applicable to appearance- and condition-specific concerns. It is intended that the devised framework for psychological adjustment to CL/P is both comprehensive and testable in future research, and that it will provide guidance for monitoring outcomes and interventions for this population.

The CL/P framework was also guided by and has built upon the existing ARC framework for visible difference (see Figure 3). While many of the constructs identified by the ARC model were included in the CL/P framework as the result of the work presented in this thesis, the latter also includes a number of additional constructs. This is likely a consequence of the largely qualitative approach underpinning the publications presented here. The results of my work also highlight the need to consider not only the aspects of adjustment which may be specific to a given condition, or subset of conditions, in the totality of an individual's experience, but also to capture appearance-related concerns which may apply to the general population. In moving forward, rather than focusing purely on the visible difference, appearance research needs to address how constructs in the fields of general body image and visible difference may overlap and interact, and to explore further the similarities and discrepancies which may exist between different visible conditions. For example, how might adjustment differ between conditions which are congenital and those which are acquired later in life? How might adjustment to a craniofacial condition differ from adjustment to a skin condition? Addressing some of these questions in the context of wider literature may help to build our conceptual understanding of psychological adjustment to appearance concerns, and help to distinguish between those concerns which are considered 'normative' and those which relate specifically to different appearance-altering conditions.

Theories and models

Now that an overarching framework for psychological adjustment to CL/P has been designed and is being utilised in research and practice, the discussion of which theories and models are most appropriate for use within this population and where theories and models may fit in relation to the framework is more pertinent. One concept which is strongly relevant to the findings of this thesis and supports the argument for a shift toward positive outcomes and strengths is that of resilience. Resilience refers to a “dynamic process encompassing positive adaptation within the context of significant adversity” (Luthar et al., 2000). It is an interactive process between risk and protective factors, is sensitive to context and demographic factors, and is multifaceted and fluctuating (Luthar et al., 2000). From this perspective, CL/P can be considered a chronic stressor that places large demands on several domains of functioning and requires ongoing adjustment over long periods of time (Baker et al., 2009). Thus, resilience can be demonstrated by the ability of the individual to report positive outcomes and to “function above the norm in spite of” (or because of) the adversity (Tusaie and Dyer, 2004). This process was evident in Publications 4, 5, 6 and 7, whereby participants described their difficult experiences but, in many cases, discussed how they had/were managing to overcome these challenges and how they would not choose to change their experiences in favour of an “easier life” (Eiserman, 2001). Interestingly, resiliency can also be described as a “more discrete personal attribute which does not presuppose exposure to substantial adversity”. From this perspective, CL/P may not constitute a significant risk factor per se; rather the ability of the individual themselves to use effective coping strategies (Lazarus and Folkman, 1984) and to tap into social resources (Baker et al., 2009) may determine adjustment. This possibility was raised by Publications 2 and 3, where children with CL/P and no additional conditions were found to report scores in line with the reference group. Conceptualising resiliency as a personal attribute may also explain some of the conflicting findings in Publications 4, 5, 6 and 7, where participants reported negative, neutral and positive responses to the same concepts.

The development of resilience in response to adversity and the notion of resiliency as a personal trait therefore both offer a constructive frame to interpret the collection of work presented in this DPhil. The CL/P literature to date, the bulk of

which has focused on the challenges, difficulties and negative impacts of CL/P, has failed to provide a coherent picture of adjustment. In the context of theories of resilience and coping, the findings of this DPhil emphasise the need to investigate characteristics of resilience and/or protective factors in the context of CL/P, and in relation to the risk factors (e.g. the burden of treatment, perceived teasing, and social anxiety) associated with CL/P (see also Eiserman, 2001; Strauss, 2001). Such characteristics need to be explored transactionally, over time, and in relation to the attributes of the individual, the familial context and the wider social environment.

From time to time within the CL/P literature, theories and models have been posed (e.g. Baker et al., 2009; Berger and Dalton, 2009). While these suggestions often have merit, they are rarely followed-up, explored further or replicated using different data, making it difficult to test theories and models in the context of CL/P. A way forward could be for clinicians and researchers to think more broadly about the theories and models which could apply to their work and to use these as a framework when working clinically or when designing/interpreting research data (see Tevik and Feragen, in press for an example).

Methodological issues within this thesis

Secondary analysis

Publications 2 and 3 arose from an opportunity to work with a colleague from Norway, who had collected routine audit data from 10- and 16-year-olds with CL/P in her capacity as a Clinical Psychologist within the Oslo Cleft Lip and Palate Team over several years. While this large amount of data afforded us the opportunity to explore challenging research questions, and to explore the effects of key contributing variables such as age, gender, cleft type and the presence of additional conditions, it also put me in the challenging position of analysing data collected by someone else, and which was originally collected for a different purpose. The main difficulty in this for me was that although I endeavoured to understand which statistical analyses had been performed and why, I was not the one to perform the analysis; rather my role was to aid interpretation of the findings and to disseminate them. While I entirely trust the integrity of my colleagues, believe this approach to have made the best of the authors' various strengths, and

consider myself to have a firm grounding in quantitative approaches, it is a goal of mine to bolster my knowledge of, and confidence in statistical analysis going forward.

Choice of qualitative methods

Publications 4, 5, 6 and 7 utilised a patient-led, qualitative approach to data collection, and a data-driven, descriptive and thematic approach to analysis. Alternative qualitative approaches to data acquisition and analysis in health research include Narrative Analysis, which views data in a story form and discusses how events are selected, organised and connected to create meaning (see Reissman, 2005), Interpretative Phenomenological Analysis (IPA), which offers contextual and subjective insight into a given phenomenon (see Smith et al., 2009) and Grounded Theory, which is designed to generate or 'discover' a theory from the data collected (see Glaser and Strauss, 2009). All three types of analysis are frequently used in studies of illness experience (Smith, 2011; Stephens, 2011) and as a method of 'giving voice' to patients (Larkin et al., 2006), which typically involves some degree of interpretation and/or postulations of theory. Thus, a case could be made for the use of one or more of these approaches with the data presented in this thesis.

Nonetheless, the primary aims of the thesis were not to explore patient experiences in-depth, to derive detailed meaning or interpretation, or to evoke theoretical discussion. Rather, the aims were to provide a broad overview of under-examined topics from the patient perspective and to offer pragmatic suggestions for future research and clinical practice. More specifically, detailed interpretation such as that offered via Narrative analysis or IPA was not considered possible for Publications 5, 6 and 7, due to the volume of data collected, while Grounded Theory was not considered appropriate due to the large number of topics covered in the interviews and the wide-ranging nature of the issues discussed. Thus, much like the broad framework approach to conceptualising CL/P, the research described within this thesis was designed to provide a platform for more detailed qualitative analysis in the future, to inform quantitative work, and to offer a broad structure for psychological intervention. However, in hindsight, Narrative analysis or IPA may have been a better fit for

Publication 4, due to the smaller sample size and the 'storytelling' nature of the data collected. When collecting the data for Publication 4, I was under time pressure and thus chose a method I felt comfortable in using, rather than the method which arguably best suited the research question. This is an important learning outcome for me and will be a salient consideration for future research projects.

Saturation

In contrast to quantitative studies in which the question of sample size can be addressed relatively simply by calculations of power, the sample size of a qualitative study is often determined by 'saturation'; the point at which no 'new' information is collected. A number of factors can influence how and when saturation is achieved, including the two arguably most important factors, the aims of the study and the heterogeneity of the population (Morse, 2000). As previously discussed, the aims of the research were to provide a broad descriptive overview of the issues important to formerly under-researched populations. Thus, the interviews were not designed to explore participants' experiences in a huge amount of detail. The heterogeneity of the samples was large in some respects (for example, with respect to age range), but not in others (for example, most participants identified as 'White British' and were considered to be of 'middle class'). Thus, saturation could be deemed to be acceptable. In the case of Publication 4, time restraints were apparent and therefore it is possible that saturation would have been more justifiable had more participants been sought; however, there was clear commonality between the participants who were interviewed and this study provided an important first step in an otherwise scarce evidence base. For Publications 5, 6 and 7, the amount of data already collected was becoming overwhelming and thus collecting additional data felt counterproductive. The decision was made to stop at this point as we believed the aims of the study had been met. Nonetheless, it must be acknowledged that the samples obtained for these studies were self-selecting; a challenge for most research and one which could affect true saturation of the research topic.

Reflexivity and triangulation

'Reflexivity' refers to the continuing self-awareness and critical self-reflection of the researcher(s), both within the context of the research itself and in relation to the dynamics observed between the researcher(s) and the participants (Finlay and Gough, 2008). It allows researchers to acknowledge their role in the development of the findings and provides a means of creating greater transparency and quality within research (Finlay and Gough, 2008).

In their comprehensive guidelines for qualitative research, Cohen and Crabtree (2006) suggested three key steps to fostering reflexivity within research. First, the researcher(s) should keep a reflexive journal. In the case of the work presented in this thesis, a detailed log of ideas, reflections and methodological decisions was kept for each study. Analysis was also seen as recursive; notes pertaining to possible codes and themes were made throughout the interviewing process and additional interview questions were included where appropriate. In addition, I discussed the methodological approach and interview experience with the supervisory team between interviews, as part of my reflective journey. Second, Cohen and Crabtree proposed that research perspectives, positions, values and beliefs should be documented in publications. This is not often seen within the field of CL/P, which is only just becoming accepting of qualitative research itself (Nelson, 2009). Although I aimed to be reflective while conducting this research and throughout this DPhil commentary, such reflections were not clearly documented within the presented publications themselves. This is a consideration which I will take forward into future projects. Finally, research should include multiple investigators, which is also a form of triangulation. In the case of the work presented in this thesis, I sought to involve clinical psychologists where possible, either directly in terms of authorship, or indirectly as ad-hoc advisors. Additionally, I discussed the research design and the research findings with the Cleft and Craniofacial Anomalies Clinical Studies Group (CSG, including patients and clinicians from various disciplines), with the research participants themselves, and with patient representatives from charitable organisations. This was done to ensure the research questions were of clinical relevance, and to gauge further opinion on the accuracy and representativeness of the findings. I believe strongly in this approach for all research and will continue to build upon what I have learned during the writing of this thesis in future work.

'Triangulation' can also refer to the application of several research methods in the study of the same phenomenon. Although the work presented in this DPhil thesis did not utilise this approach in full, my plan in moving forward is to build upon the work presented here in both a qualitative and quantitative capacity. Specifically, quantitative approaches can be designed in light of the qualitative findings presented in this thesis, and qualitative work can be used to support and/or explore the quantitative findings of this thesis in more detail. More broadly, the presented work can inform the methodological design of future research and potential intervention studies. In addition, the use of more creative methods of data collection is of interest to me for future projects, for example, the use of participatory activities with children (see Darbyshire et al., 2005).

Focus on neglected groups

For this DPhil thesis, I chose to present publications that investigated and gave voice to some professionally 'neglected' groups over others. This was partly strategic; some 'hard-to-access' groups are in fact easier to access than others, particularly in light of my growing relationships with relevant and representative organisations. Additionally, some of the analysis was based on data which had already been collected, such as in the case of Publications 2 and 3. Nonetheless, this approach meant that some of the neglected groups which were identified in Publication 1 were not discussed in further detail within this thesis, except briefly within the articles themselves. Most notably, this includes minority ethnic groups, other 'social groupings' (including those reporting low socioeconomic status, non-English speaking families, and those from less developed countries) and the needs of the wider family (including, for example, siblings and grandparents). Since the completion of the publications presented in this DPhil, I have carried out a collaborative qualitative study with the Cleft Lip and Palate Association, to assess the information and support needs of unaffected siblings of children with CL/P. This publication has implications for the inclusion of siblings within cleft care and the wider CL/P community, and CLAPA are now looking into developing activities and events for siblings in collaboration with another UK charity; an endeavour which will likely become part of an ongoing service evaluation carried out by the Centre for Appearance Research. As part of my ongoing relationship with CLAPA, potential studies involving grandparents and those from minority

ethnic groups are also planned for future years. Additionally, the work presented in this thesis has played a key role in informing the development of a nationwide cohort study entitled The Cleft Collective (see the 'Current work' section below for more information). Within the Cohort Studies, we have made initial efforts to recruit families who are non-English speaking, from minority ethnic communities and/or who report a low socioeconomic status, and are continually reviewing and evaluating both our approach and the representativeness of the data we are collecting. Finally, I am part of the team leading the Global Holistic Outcomes Task Force for Cleft and Craniofacial Anomalies (see 'Future plans' section below for more information), whose aim is to increase awareness of the psychological impact of CL/P among clinicians working in less developed countries around the world, and to begin to implement basic measurement of the psychological aspects of CL/P into these teams.

Overlap between disciplines

A final issue which was raised in Publication 1 and was beyond the scope of the current thesis was the current lack of interdisciplinary research, in spite of the multidisciplinary nature of CL/P treatment and despite cleft services being centralised in the UK.

One key association which currently lacks exploration is the overlap between psychological adjustment and speech and language difficulties. While pioneering research suggests that the social challenge of a visible difference can be overcome by a good level of social interaction skills (Rumsey and Bull, 1986; Rumsey, Bull and Gahagan, 1986), this could be jeopardised in the case of CL/P where an audible difference may also be present (Sell, 2005). In light of these considerations, The Cleft Collective Cohort Studies are collecting a wide range of data pertaining to both psychological wellbeing and speech and language development, with the aim of investigating the interaction between these two key variables and the implications for cleft care. In addition, I was recently invited to contribute to the writing of a collaborative article between psychologists and speech and language therapists, which served as a good introduction to the importance and potential of such interdisciplinary work (please see Bibliography section).

It is hoped that The Cleft Collective Cohort Studies, among other research projects, will also allow for investigation of the overlap between psychological adjustment and other key disciplines involved in cleft care, including, for example, the impact of hearing difficulties and the experience of surgery and treatment.

Current work

The aim of this section is to provide an overview of my current work in the field, and to describe how I am utilising the findings of this DPhil to guide future research and inform clinical practice. The section outlines my work for The Cleft Collective, the largest cleft lip and palate research programme in the world to date, as well as my growing investment in Patient and Public Involvement and ongoing collaborations with other stakeholders in the field. Examples of my other professional activities are also included.

The Cleft Collective

In September 2011 I began contributing to the development of a new research programme entitled The Cleft Collective (www.cleftcollective.org.uk), an initiative of the UK charity, the Healing Foundation. In March 2012 I joined the programme, based at Bristol University, as a full-time Research Associate for five years. Alongside the Cleft Clinical Trials Unit, based at Manchester University, we have established two parallel cohort studies (a birth cohort and a five-year-old cohort). We are collecting biological samples, including blood and tissue from the child and saliva from parents and siblings, as well as comprehensive questionnaire data pertaining to environmental factors and psychological wellbeing from parents around the time of their child's CL/P diagnosis. We then hope to follow the progress and development of the enrolled families over time, with the aim of answering three key questions that parents often ask: 1) What caused my child's cleft? 2) What are the best treatments for my child? 3) Will my child be OK? The Cleft Collective Cohort Studies will build on and complement existing cohort studies within the general population (see www.bristol.ac.uk/alspac), as well as past and current outcomes studies in Europe (Eurocleft; see Shaw et al., 2001) and the United States (Americleft, see Long et al., 2011). My role, as well as contributing to the overall research programme, is to lead the psychological strand

of the research (question 3) and to represent the patient voice through ongoing engagement in Patient and Public Involvement (PPI) activities.

Establishing the cohort studies has been an extremely challenging process. The number, diversity and geographical spread of stakeholders has highlighted a range of competing needs, agendas and priorities. Extensive and ongoing consultation and negotiation with each stakeholder is required, as is the need to be flexible and supportive. In addition, and as previously discussed, deciding which constructs to measure, how to measure them and at which time point represented a substantial challenge, which was overcome only through extensive collaboration with the SIGs and other expert groups from the beginning of the project.

Nonetheless, this process has been enormously rewarding. As well as helping me to develop networks with patients, multidisciplinary cleft teams and charities, and to begin to build esteem in this field, my work for The Cleft Collective has led to the opportunity to collect standardised psychology data from a very large sample, across disciplines and across locations. The cohort study will collect longitudinal, prospective data from several members of each family that participates, regardless of the type of cleft diagnosis, and at key points during the child's developmental trajectory. If successful, this will represent a ground-breaking achievement in CL/P research, with the potential to be world-leading in a number of key research areas. In addition, psychologists working in cleft teams around the UK have recently agreed to adopt the same pack of standardised measures for their five-year national audit, which has required a reorganisation for several years. This will allow psychology data to be compared across sites and to be included in national standardised databases such as the CRANE database (www.crane-database.org.uk). My work for The Cleft Collective thus far has also led to the writing of two collaborative research papers, one of which details the opportunities and challenges of setting up a cohort study (under review) and one of which describes the process of developing a conceptual framework and choosing appropriate measures to inform future research and audit (in preparation). To promote the research and the reputation of the universities involved I have given high-profile presentations to a number of potential funders, as well as to HRH Countess of Wessex in her role as Patron to the Healing Foundation. Finally, and most rewardingly, much of the work presented in this doctoral thesis has been

influenced by, and fed into, The Cleft Collective research programme. In February 2014 I was promoted to Research Fellow as a permanent (funding-based) member of staff, in recognition of my work to date.

Patient and Public Involvement (PPI)

Until relatively recently, the philosophy that ‘doctor knows best’ took precedence and research was dictated by governing bodies and researchers themselves. Today, there is a growing ethos of patient autonomy and involvement, where patients have a much larger say in their own treatment and in setting priorities for research. During the development of The Cleft Collective research programme, I ran several PPI workshops between 2011 and 2012. Here, we discussed the meaning of PPI with participants and the ways in which they might become involved with the research programme. At these workshops, and through ongoing contact with PPI representatives, patients have contributed to the design of the cohort studies and the materials which are given to participating families (including information sheets, leaflets and questionnaires). I also established a confidential database of patients who are interested in participating in research and PPI activities, and set up a ‘Cleft Image Bank’, to which individuals with CL/P and their families can contribute photographs of their journey. Significant improvements have been made to The Cleft Collective research programme as a result of this input, for which I am extremely grateful. I have since advocated the use of PPI in research at a number of conferences and events, and have attended events hosted by the leading PPI advisory group, INVOLVE (www.invo.org.uk). On an individual study level, PPI has also contributed to the development, execution, analysis and dissemination of many of the publications presented in this doctoral thesis.

I view PPI as not only involving patients, but other stakeholders as well, including (for example) the Psychology SIG, members of the cleft teams and representative organisations such as CLAPA. This ongoing negotiation allows for clear communication of joint objectives and collaboration to overcome any difficulties, and prevents stakeholders from working in the silos of their respective disciplines or fields of expertise. I believe PPI to be crucial to the implementation, impact and

sustainability of research findings into practice, both in the field of CL/P and within the wider research community.

Collaboration

Throughout this journey, I have been very fortunate to meet and work alongside a number of like-minded people with similar ideas and ambitions for the research field. At the top of this list is Dr Kristin Billaud Feragen, Clinical Psychologist for the Cleft Lip and Palate Team in Oslo, Norway. As well as the four joint publications presented within this thesis (Publications 2, 3, 5 and 7), Dr Feragen and I have collaborated on a further four papers (currently in press/under review) and four conference presentations. Three of these papers involved the comprehensive analysis of the psychological adjustment of children with CL/P at age 10 and age 16, as well as longitudinal analysis of adjustment from age 10 to age 16. The fourth collaboration described an investigation of potential associations between speech, language, reading and psychological variables, and involved two speech and language therapists from the Oslo cleft team. Additional future collaborations are also planned/in progress.

I also enjoy a close working relationship with the Cleft Lip and Palate Association (CLAPA), the only UK-wide organisation specifically devoted to supporting those with and affected by CL/P. I supervise and run the independent evaluation of their regional services, producing professional reports each year which allow them to develop their services and apply for further external funding. I am also involved in several of their committees, including the Adult Voices Council, the Children and Young People's Council, the Bristol and South West Branch, and the Regional Coordinators Advisory Panel. On an ad-hoc basis, I provide ongoing consultancy work for CLAPA, including input into their annual membership surveys and external grant applications. The relationship with CLAPA has helped me to recruit participants into studies and PPI activities, promote The Cleft Collective research programme, disseminate my research findings, feed my knowledge and ideas into practice and provide CLAPA with an evidence base from which to work. The relationship is extremely beneficial for both parties, and a number of future collaborations are planned, including joint funding applications for a number of small research projects.

Being able to travel for work has also provided me with opportunities to forge ongoing relationships with researchers, clinicians and charities abroad. In particular, I have enjoyed working with members of the Social Sciences department at the University of Buenos Aires in Argentina. As well as delivering two presentations to the department, I was able to meet with the Minister for Health and visit the local CL/P team to share current practice and develop future proposals. We have since presented a collaborative study at the 9th Hispanic Latin American Congress of Eating Disorders and members of the University of Buenos Aires are now involved in the European Cooperation in Science and Technology (COST) Action (see below).

In 2014 I also met Dr John Thompson, a Senior Research Fellow in Paediatrics at the University of Auckland in New Zealand. After visiting the UK to find out more about our research in CL/P, John was awarded a significant grant to replicate the original UK CSAG study in New Zealand over the next four years. John is keen to remain in contact and to collaborate on a number of joint projects and publications in the near future.

Another colleague from New Zealand, Kenny Ardouin, who was born with CL/P himself and now directs the leading charity for CL/P in New Zealand (Face It NZ), contacted me after seeing a live online broadcast of my presentation at the CLAPA conference in 2013. He asked if I would give a similar presentation using Skype technology to a large group of children affected by CL/P at the charity's first Youth Camp in Auckland. I delivered this presentation, after adapting it for a younger audience, and also provided Kenny with some hand-outs for the children to take home. I have since been invited to give the same talk again at the Youth Camp in 2015, and to speak to parents, patients and clinicians at the charity's Annual General Meeting in the same year. Psychological support is scarce in New Zealand, and so the opportunity to discuss psychological issues and to hear about other people's experiences through the research was fed back as being "emotive, informative and essential". This spring, Kenny visited the Centre for Appearance Research and The Cleft Collective, which was extremely interesting and inspiring for all involved. It is a pleasure to stay in contact with Kenny and the charity, and I hope to be involved in additional collaborations in the future.

Examples of additional professional activities

In addition to the work described above, I have also engaged in a number of teaching activities. I have led and co-directed several lectures and seminars for students enrolled on the following courses at UWE: BSc Psychology (Years 2 and 3); MSc Health Psychology; and MSc Research Methods in Psychology. I have also delivered two invited specialist seminars to students attending the School of Oral and Dental Sciences at the University of Bristol. Finally, I was invited to lead a seminar to a varied professional audience working on the Avon Longitudinal Study of Parents And Children (ALSPAC), a prominent longitudinal cohort study in the UK.

To date I have contributed to three successful funding applications, including: Big Lottery Reaching Communities bid in collaboration with the Cleft Lip and Palate Association (awarded £284,881 and £199,838); and UWE QR funded workload bundles for academic/research development (awarded £3,000). I am currently involved in the development of two further funding bids. In addition I have contributed to three pitches to potential funders which have thus far secured £1.4 million in funding for a National cleft lip and palate research programme. I have played a part in the preparation of two PhD funding bids, as well as an interdisciplinary collaborative Programme Grant (NIHR) and two further funding bids in collaboration with two external charities.

Over the last two years I have attended several relevant courses aimed at continuing my professional development. These courses have included: Systematic Reviews and Meta-Analysis; Genetics and Genomics; Managing the Media; Good Clinical Practice (GCP); Qualitative methodology and analysis; and relevant conference attendance (including Health and Clinical Psychology, Appearance, Cleft Lip and Palate, Dental and Oral Sciences, Core Outcomes and Patient and Public Involvement).

I am a member of several professional bodies, including: the British Psychological Society (postgraduate member); Cleft Lip and Palate Association (member and research consultant); Psychology Special Interest Group for Cleft and Craniofacial Anomalies (member and research contributor); American Cleft Palate-Craniofacial Association (member and peer-reviewer); Cleft New Zealand (member and contributor).

Finally, I am a peer-reviewer for four reputable international academic journals, including: the Cleft Palate-Craniofacial Journal; the Journal of Clinical Child Psychology and Psychiatry; Body Image; and the Journal of Paediatrics and Child Health.

Future plans

The aim of this final section is to outline a number of ambitions for the future of CL/P research and my own professional development.

In a recent priority setting initiative by the James Lind Alliance (www.lindalliance.org), clinicians, researchers, individuals born with cleft and their families voted psychological intervention and long-term outcomes to be the two most important unanswered research questions in CL/P. The Cleft Collective research programme will put the UK in a unique and privileged position to be able to answer some of these 'big' research questions, and I hope to be at the forefront of this vital work. In particular, I feel further investigation of psychosocial interventions for the field of CL/P is crucial. Over time, The Cleft Collective studies will provide the foundation for a huge resource of information about children with CL/P and their families. This resource will be available to clinicians and researchers both within and outside of the UK to use for ethically approved projects, and I hope to be involved in some important national and international collaborations. All of our research findings will be shared widely, and I would like to play a key part in incorporating these findings into clinical practice around the world.

In connection with this, the Centre for Appearance Research recently established a large COST Action (IS1210) to tackle the physical and psychosocial consequences of dissatisfaction with appearance. The Action aims to “co-ordinate and increase research across Europe, offer support to the high proportion of female and early career researchers in this field and...forge crucial links between researchers, practitioners and policy makers, offering the potential for significant benefits to the millions of Europeans adversely affected by [appearance concerns]”. I attended a conference in December 2014 to find out more about the COST network and how I may be able to contribute to the Action.

I would also enjoy a role in the expansion of research and practice across Europe in relation to CL/P specifically. I am contributing to the development and delivery of a psychosocial research symposium at the 10th European Craniofacial Congress in Sweden in 2015. I will also be contributing to the design and delivery of an international psychosocial training day for health professionals involved in the care of children with craniofacial conditions at the 13th International Cleft Congress in Chennai, India, in 2017. Finally, and building on the work of previous task force activities (see Broder 2014; Semb, 2014), I am part of the team leading the International Holistic Outcomes Task Force for Cleft and Craniofacial Anomalies. We aim to employ a tiered approach to the implementation and measurement of psychological care for patients, which can be achieved by all participating countries regardless of cultural issues and the availability of resources. The knowledge attained through this DPhil will be disseminated and expanded upon during each of these key reputable international events.

The South West Cleft Team, previously based at Frenchay Hospital, has recently moved to the University of Bristol Dental School where The Cleft Collective research programme is based. I hope this will allow me the opportunity to become more integrated into the cleft team, and to assist with the execution of the research and the implementation of the findings.

On a personal level, I thoroughly enjoy working for the university and the supportive and creative space it provides. Over the next few years I hope to be deserving of a Senior Research Fellow post and/or the recipient of a postdoctoral fellowship. My long-term aspiration is to establish my own research group in the field of cleft and craniofacial research, within the supportive and thriving environment of CAR. I would also enjoy feeding some of this knowledge and expertise back into the university via additional teaching and further supervision of students and less experienced colleagues. On a wider scale, I feel relatively little is understood about participation in applied research and the benefits it can have. It is my ambition to help to create a research 'community', in which taking part in research and clinical audit is a normal and enjoyable part of the treatment pathway, and in which patients can clearly see and benefit from the impact of their contribution.

Conclusions

Collectively, the findings presented in this DPhil imply that a conceptual and methodological shift is required in both academic research and clinical practice in the field of CL/P. CL/P is a lifelong condition, the psychological adjustment to which is influenced by a range of interacting factors and processes. Individual variation in adjustment is considerable, as are fluctuations in adjustment over time and across situations. Rather than using an approach which is inherently pathologising, and/or in which the cleft itself is perceived to be a risk factor for poor outcomes, it may instead be more productive to conceptualise the cleft and its treatment as an underlying stressor which is present throughout life (Lansdown et al., 1997). While this stressor is likely to make continuous calls on energy reserves and coping resources, the same 'normal' developmental stages and life events experienced by the general population also apply. These life stages and events have the potential to compound research findings and should thus be accounted for. To prevent the cleft becoming a 'hook' on which distress resulting from other sources is hung, an appreciation of the wider context and broader experiences of the individual is essential. A holistic and systemic approach, which encompasses previously neglected subgroups of patients is also crucial, as is the involvement of patients and families in setting research agendas and improving service delivery.

Agreement upon the key components and characteristics contributing to a 'positive psychological outcome', as well as how and when to measure the contributing factors and processes consistently, is vital to the future of this field. An increased effort to obtain large samples would reduce the potential for misleading or inconclusive results, and allow for the investigation of clinically important subgroups and potentially interacting variables. An increase in qualitative research is needed, as is an appreciation and integration of the patient perspective as a whole. The implementation of longitudinal research is challenging and requires a long-term investment, but is the best chance of answering some of the 'big' outstanding research questions, such as those relating to the optimal type and timing of interventions, as well as a better understanding of the longer-term outcomes for those affected by CL/P.

By shifting our conceptual and methodological approach to the study and care of individuals affected by CL/P and their families, we may begin to close some of the

gaps in our understanding of psychological adjustment to this relatively common condition. Specifically, a steer towards an approach which is holistic rather than narrow, inclusive rather than exclusive, normalised rather than pathologised, appreciative of the patient perspective and encompassing of strengths is required.

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Appendices

Appendix 1

Map of evidence against university doctoral descriptors

The award of a Doctorate of the University (other than a Higher Doctorate) requires that a candidate should demonstrate that he/she:

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| <p><i>(i) has conducted enquiry leading to the creation and interpretation of new knowledge through original research or other advanced scholarship, shown by satisfying scholarly review by accomplished and recognised scholars in the field</i></p> | <p>Six peer-reviewed journal articles have been presented.</p> <p>These publications are supplemented by evidence of the author's impact on the wider research field, provided in the Appendices.</p> |
| <p><i>(ii) can demonstrate a critical understanding of the current state of knowledge in that field of theory and/or practice</i></p> | <p>This thesis was based upon a comprehensive literature review and a critical appraisal of the existing gaps in the general knowledge base, which was published in a seminal book in this field.</p> <p>In addition, each publication and its commentary offers a critique of the state of knowledge in relation to the relevant topic areas.</p> |
| <p><i>(iii) shows the ability to conceptualise, design and implement a project for the generation of new knowledge at the forefront of the discipline or field of practice including the capacity to adjust the project design in the light of emergent issues and understandings</i></p> | <p>The author contributed significantly to the conceptualisation, design and implementation of two of the six peer-reviewed publications presented (Publications 2 and 3).</p> <p>The author initiated, designed and implemented the remaining four publications (4, 5, 6 and 7) with minimal supervision and in collaboration with two</p> |

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| | <p>other leading authors in this field.</p> <p>All six of the publications underwent some degree of adjustment in response to emerging issues, including methodological alterations and the reorganisation of concepts and data presentation.</p> |
| <p><i>(iv) can demonstrate a critical understanding of the methodology of enquiry</i></p> | <p>This thesis is based largely on a critique of current methodologies in the field and the contribution of related challenges and limitations to the conflicted evidence base.</p> <p>In addition, each publication and its commentary provide a critique of methodology specific to that subject area.</p> <p>Finally, each publication provides discussion of alternative methodological approaches which may help to alleviate some of the contradictory findings within the literature.</p> |
| <p><i>(v) has developed independent judgement of issues and ideas in the field of research and/or practice and is able to communicate and justify that judgement to appropriate audiences</i></p> | <p>Many of the ideas presented in this thesis challenge current thinking in this field. The publications and the accompanying commentary critique current approaches to research and practice and suggest alternatives.</p> <p>The dissemination of these findings has required the author to adjust their presentation according to a wide range of audiences. This is also demonstrated by additional evidence in the Appendices.</p> |

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| <p><i>(vi) can critically reflect on his/her work and evaluate its strengths and weaknesses including understanding validation procedures</i></p> | <p>Each publication includes a reflection of the author's work, including strengths and limitations, as well as suggestions for improvements in future research.</p> <p>In addition, the commentary provided within this thesis has reflected on the author's journey and demonstrated the progression of personal and professional growth.</p> |
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Appendix 2

Bibliography

Peer-reviewed publications

Stock, N.M., Whale, K., Jenkinson, E., Rumsey, N. and Fox, F. (2013) Young people's perceptions of visible difference. *Diversity and Equality in Health and Care*, 10(1), pp. 41-51.

Stock, N.M. and Rumsey, N. (2015, e-pub 2013) Parenting a child with a cleft: The father's perspective. *Cleft Palate-Craniofacial Journal*. 52(1), pp. 31-43.

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Norman, A., Persson, M., **Stock, N.M.**, Rumsey, N., Sandy, J., Waylen, A., Edwards, Z., Hammond, V., Partridge, L. and Ness, A. (2015, e-pub 2014) The effectiveness of psychosocial intervention for individuals with cleft lip and/or palate and their parents: A systematic review. *Cleft Palate-Craniofacial Journal*, 52(3), pp. 301-310. doi: <http://dx.doi.org/10.1597/13-276>.

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Stock, N.M., Humphries, K., St Pourcain, B., Bailey, M., Persson, M., Ho, K.M., Ring, S., Marsh, C., Albery, L., Rumsey, N. and Sandy, J. (e-pub ahead of print, 2015) Opportunities and challenges in establishing a cohort study: An example from cleft lip/palate research in the UK. *Cleft Palate-Craniofacial Journal*.

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Book chapters

Rumsey, N. and **Stock, N.M.** (2013) Living with a cleft: Psychological challenges, support and intervention. In: Berkowitz, S., ed. (2013) *Cleft Lip and Palate: Diagnosis and Management, 3rd edition*. Berlin Heidelberg: Springer-Verlag, pp. 907-915.

Invited articles

Stock, N.M. (2012) Appearance Matters - and so does research: Reflections on becoming a researcher. *PsyPAG Quarterly*. 78, pp. 33-34.

Stock, N.M. (2013) Not such a quick fix: Defining the challenges and myths associated with disfigurement. *Journal of Aesthetic Nursing*, 2(6), pp. 302-303.

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Professional reports

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Stock, N.M., Whale, K., Jenkinson, E., Fox, F. and Rumsey, N. (2011) *An evaluation of a school-based intervention to promote positive attitudes and behaviours towards people with facial disfigurement: Main study report*. Centre for Appearance Research: University of the West of England, Bristol. February 2011.

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Stoneman, K., **Stock, N.M.**, Cunniffe, C., Martindale, A. and Rumsey, N. (2015). *CLAPA Regional Coordinators Project: Evaluation Interim Report. Focus group studies in the South East UK region*. Centre for Appearance Research: University of the West of England, Bristol. March 2015.

Appendix 3

National and international conference presentations

Oral presentations

Stock, N.M. (2010) *Body image and exercise*. Bristol, UK, 02 November 2010. University of the West of England: Psychology postgraduate conference.

Stock, N.M. and Rumsey, N. (2011) *Psychosocial issues in cleft lip and palate*. York, UK, 14 April 2011. Annual Scientific Conference: Craniofacial Society of Great Britain and Ireland [winner of the President's Medal].

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Jenkinson, E., **Stock, N.M.**, Thomas, E. and Clarke, A. (2012) *Do psychosocial specialists find CBT an acceptable approach to working therapeutically with people with appearance-altering conditions or injury?* Bristol, UK, 04 July 2012. Appearance Matters 5: Biannual conference of the Centre for Appearance Research.

Townsend, D. and **Stock, N.M.** (2012) *Exploring media representations of exercise in women's magazines*. Bristol, UK, 03 July 2012. Appearance Matters 5: Biannual conference of the Centre for Appearance Research.

Stock, N.M., Cunniffe, C., Jenkinson, E., Preston, R. and Rumsey, N. (2012) *Local support: Needs, opportunities and outcomes in cleft lip and palate*. Bristol,

UK, 03 July 2012. Appearance Matters 5: Biannual conference of the Centre for Appearance Research.

Stock, N.M. (2012) *The Cleft Collective: Patient and Public Involvement: How you can get involved*. Bristol, UK, 08 September 2012. The Annual Conference of the Cleft Lip and Palate Association.

Feragen, K.J.B. and **Stock, N.M.** (2013) *How inclusion and exclusion criteria may affect outcome measures in cleft lip and palate: An example from psychological research*. Birmingham, UK, 18 April 2013. Annual Scientific Conference: Craniofacial Society of Great Britain and Ireland.

Humphries, K., **Stock, N.M.** and The Cleft Collective Cohort Studies Team (2013) *The Cleft Collective Cohort Studies*. Birmingham, UK, 18 April 2013. Annual Scientific Conference: Craniofacial Society of Great Britain and Ireland.

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Tierney, S. and **Stock, N.M.** (2013) *Conducting 'good' research: Why it's important to have Patient and Public Involvement*. Glasgow, UK, 05 October 2013. The Annual Conference of the Cleft Lip and Palate Association.

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Stock, N.M., Dixson-Smith, A., Ruel, T. and Rumsey, N. (2014) *Adult voices: the support needs of adults with cleft lip/palate and implications for practice*. Bristol, UK, 02 July 2014. Appearance Matters 6: Biannual conference of the Centre for Appearance Research.

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Bamford, J., Blyth, K., Brown, S., Kelly, T. and **Stock, N.M.** (2015). *A validated patient-reported outcome measure for satisfaction with hearing, appearance and speech in the cleft lip and palate population: The Cleft Hearing, Appearance and Speech Questionnaire (CHASQ)*. London, UK, 16 April 2015. Annual Scientific Conference: Craniofacial Society of Great Britain and Ireland.

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Stock, N.M. (2015) *"It doesn't all just stop at 18". Long-term outcomes in cleft lip/palate and implications for practice.* Gothenburg, Sweden, 26 June 2015. 10th European Craniofacial Congress.

Stock, N.M. (2015) Achieving consensus in outcome measurement: An example from cleft lip/palate research in the UK. London, UK, 17 September 2015. Division of Health Psychology annual conference.

Invited talks

Stock, N.M. (2011) *It's not just about the surgery: Factors influencing psychosocial outcomes in clefting and how to measure them.* Bristol, UK, 02 November 2011. University of Bristol, School of Oral and Dental Sciences Seminar Series.

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Stock, N.M. (2012) *Satisfaction with appearance: The role of magazine consumption in appearance beliefs and behaviours.* Buenos Aires, AR, 19 October 2012. Department of Psychology and Health: University of Buenos Aires.

Humphries, K. and **Stock, N.M.** (2013) *The Cleft Collective Gene Bank and Cohort Studies.* Bristol, UK, 23 October 2013. Presentation to HRH The Countess of Wessex in her role as Patron of the Healing Foundation.

Stock, N.M. (2013) *Does appearance matter? The Psychology of satisfaction with appearance.* Auckland, NZ, 13 December 2013. Youth Camp 2013, Cleft New Zealand.

Stock, N.M. and Sandy, J.R. (2014) *'One born every three minutes'. Researching cleft lip/palate in the UK and beyond.* London, UK, 30 January 2015. The Healing Foundation, Royal College of Surgeons.

Stock, N.M. (2015) *Does appearance matter? The psychology of satisfaction with appearance.* Auckland, NZ, 10 April 2015. Youth Camp 2015, Cleft New Zealand.

Stock, N.M. (2015) *Psychosocial adjustment to cleft lip/palate: Examples from research in the UK.* Auckland, NZ, 26 May 2015. Annual General Meeting 2015, Cleft New Zealand.

Stock, N.M. and Davies, A. (2015) *The Cleft Collective: Where are we now?* Bristol, UK, 1 June 2015. The Avon Longitudinal Study of Parents and Children.

Stock, N.M. (2015) *Achieving consensus in outcome measurement [Psychology Symposium: Joining up research and practice: the benefits of collaboration].* Gothenburg, SWE, 25 June 2015. The 10th European Craniofacial Congress.

Poster and e-poster presentations

Stock, N.M. and Rumsey, N. (2013) *Parenting a child with a cleft: The father's perspective.* Orlando, FL, 05-10 May 2013. The 12th International Congress on Cleft Lip/Palate and Related Craniofacial Anomalies [e-poster].

Blyth, K., Bamford, J., Brown, S., Kelly, T. and **Stock, N.M.** (2014) *A patient and carer reported outcome measure for satisfaction with appearance in the cleft lip and palate population.* Bristol, UK, 01-02 July 2014. Appearance Matters 6: Biannual conference of the Centre for Appearance Research [poster].

Tierney, S., **Stock, N.M.**, Preston, R. and Cunniffe, C. (2014) *Stronger together: Collective endeavours of a cleft research organisation to promote the role of young people affected by cleft in research.* Birmingham, UK, 26-27 November 2014. INVOLVE 2014: Changing Landscapes [poster].

Stoneman, K., **Stock, N.M.**, Cunniffe, C. and Rumsey, N. (2015) *The psychosocial impact of cleft lip and/or palate on unaffected siblings.* London, UK, 15-17 April 2015. Annual Scientific Conference: Craniofacial Society of Great Britain and Ireland [poster].

Appendix 4

Additional presentations

Stock, N.M. (2011) *Psychosocial issues in cleft lip and palate: Findings of a literature review*. London, UK, 11 January 2011. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. and Rumsey, N. (2011) *Psychosocial issues in cleft lip and palate: A recap*. London, UK, 22 June 2011. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. (2011) *A proposed framework for cleft lip and palate research in the UK*. London, UK, 22 June 2011. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. (2011) *Measuring satisfaction with appearance*. Birmingham, UK, 07 September 2011. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. (2012) *Psychology measures: an update*. London, UK, 11 January 2012. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. and Rumsey, N. (2012) *The Cleft Collective: Birth Cohort and Gene Bank Study*. Bristol, UK, 25 April 2012. Cleft and Craniofacial Nursing Special Interest Group.

Stock, N.M. (2012) *The Cleft Collective: Update and choosing Psychological measures*. Bristol, UK, 25 April 2012. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. (2012) *The Cleft Collective: Draft protocols and measures*. Manchester, UK, 29 June 2012. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. (2012) *The Cleft Collective: Plans and Progress*. Bristol, UK, 26 July 2012. The Healing Foundation Advisory Panel.

Stock, N.M. and Rumsey, N. (2012) *The Cleft Collective: Psychology*. Bristol, UK, 28 September 2012. The Healing Foundation Professionals Workshop.

Stock, N.M. (2013) *The Cleft Collective: Progress update and psychology measures*. London, UK, 16 January 2013. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. (2013) *The Cleft Collective: Update*. Birmingham, UK, 17 April 2013. Cleft and Craniofacial Psychology Special Interest Group. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. (2013) *The Cleft Collective: Public Involvement*. Bristol, UK, 22 April 2013. The Healing Foundation meeting for potential funders.

Stock, N.M. (2013) *The Cleft Collective Gene Bank and Cohort Studies*. Bristol, UK, 25 April 2013. South West Branch Annual General Meeting: The Cleft Lip and Palate Association.

Humphries, K. and **Stock, N.M.** (2013) *The Cleft Collective Gene Bank and Cohort Studies*. Manchester, UK, 30 April 2013. Manchester Cleft Lip and Palate Team.

Stock, N.M. (2013) *Adult Voices: Experiences and support needs of adults with CL/P*. Cambridge, UK, 19 May 2013. Regional Coordinators Project Launch Event: The Cleft Lip and Palate Association.

Stock, N.M. (2013) *Adult Voices: Experiences and support needs of adults with CL/P*. London, UK, 01 June 2013. Adult Voices national quarterly meeting: The Cleft Lip and Palate Association.

Stock, N.M. (2013) *The Cleft Collective and national audit: Progress update*. Bristol, UK, 17 June 2013. Cleft and Craniofacial Psychology Special Interest Group.

Humphries, K. and **Stock, N.M.** (2013) *The Cleft Collective Gene Bank and Cohort Studies*. Cambridge, UK, 06 August 2013. Addenbrooke's Cleft Lip and Palate Team.

Stock, N.M. and Humphries, K. (2013) *The Cleft Collective and national audit: Update*. London, UK, 30 September 2013. Cleft and Craniofacial Psychology Special Interest Group.

Humphries, K. and **Stock, N.M.** (2013) *The Cleft Collective Gene Bank and Cohort Studies*. London, UK, 08 October 2013. Annual General Meeting of the Healing Foundation.

Stock, N.M. and Stoneman, K. (2013) *Regional Coordinators Project: How can you help?* London, UK, 09 November 2013. National Volunteer Day: Cleft Lip and Palate Association.

Humphries, K., **Stock, N.M.** and Bailey, M. (2013) *The Cleft Collective Gene Bank and Cohort Studies: Training.* Birmingham, UK, 18 November 2013. Birmingham Children's Hospital, Cleft Lip and Palate Team.

Humphries, K., **Stock, N.M.** and Bailey, M. (2013) *The Cleft Collective Gene Bank and Cohort Studies: Training.* Swansea, UK, 06 December 2013. South Wales Cleft Lip and Palate Team.

Humphries, K., **Stock, N.M.** and Bailey, M. (2013) *The Cleft Collective Gene Bank and Cohort Studies: Training.* London, UK, 11 December 2013. Guy's and St. Thomas' Hospital, Cleft Lip and Palate Team.

Stock, N.M. and Rumsey, N. (2014) *The Cleft Collective and national audit: an update.* Manchester, UK, 31 January 2014. Cleft and Craniofacial Psychology Special Interest Group.

Stock, N.M. (2014) *CLAPA Regional Coordinators Project: Evaluation.* London, UK, 06 February 2014. Regional Coordinators Project Advisory Group: The Cleft Lip and Palate Association.

Humphries, K., **Stock, N.M.** and Bailey, M. (2014) *The Cleft Collective Gene Bank and Cohort Studies: Training.* Manchester, UK, 06 March 2014. Manchester Children's Hospital, Cleft Lip and Palate Team.

Humphries, K., **Stock, N.M.** and Bailey, M. (2014) *The Cleft Collective Gene Bank and Cohort Studies: Training.* Oxford, UK, 28 March 2014. Spires Centre, Cleft Lip and Palate Team.

Stock, N.M. (2014) *The Cleft Collective and national audit: an update.* Oxford, UK, 09 April 2014. Cleft and Craniofacial Psychology Special Interest Group.

Humphries, K., **Stock, N.M.** and Bailey, M. (2014) *The Cleft Collective Gene Bank and Cohort Studies: Training.* London, UK, 02 May 2014. Great Ormond Street Hospital, Cleft Lip and Palate Team.

Humphries, K., **Stock, N.M.** and Bailey, M. (2014) *The Cleft Collective Gene Bank and Cohort Studies: Training*. Nottingham, UK, 19 May 2014. Nottingham and Trent Cleft Lip and Palate Team.

Humphries, K. and **Stock, N.M.** (2014) *The Cleft Collective Gene Bank and Cohort Studies: Training*. Edinburgh, UK, 01 September 2014. Hospital for Sick Kids, Cleft Lip and Palate Team.

Humphries, K. and **Stock, N.M.** (2014) *The Cleft Collective Gene Bank and Cohort Studies: Training*. Salisbury, UK, 22 September 2014. Salisbury Hospital, Cleft Lip and Palate Team.

Humphries, K., **Stock, N.M.** and Bailey, M. (2014) *The Cleft Collective Gene Bank and Cohort Studies: Training*. Bristol, UK, 14 October 2014. South West Cleft Unit, Bristol Dental Hospital.

Humphries, K., **Stock, N.M.** and Bailey, M. (2014) *The Cleft Collective Gene Bank and Cohort Studies: Training*. Chelmsford, UK, 19 November 2014. Broomfield Hospital, Cleft Lip and Palate Team.

Humphries, K., **Stock, N.M.** and Bailey, M. (2014) *The Cleft Collective Gene Bank and Cohort Studies: Training*. Liverpool, UK, 13 January 2015. Alder Hey Children's Hospital, Cleft Lip and Palate Team.

Appendix 5

Testimonials

As part of my demonstration of my wider influence in this research field, I asked a number of colleagues to provide a short testimonial. These are presented below.

Dr Kristin Billaud Feragen

Clinical Psychologist and Post-Doctoral Researcher for the Oslo Cleft Lip and Palate Team, Norway

"I first met Nicola at a conference in 2011, where she was presenting an overview of psychosocial issues in cleft lip and palate. I perceived her presentation as engaging and of high quality, and was therefore very happy to get to know her during the conference. Later, while working on my postdoctoral fellowship at the Centre for Appearance Research (July 2012 to August 2013), working in Bristol provided the opportunity for an efficient collaboration with Nicola, which has continued since then.

My respect for Nicola has only been growing during the four years I have known her. I perceive her as devoted to her work and driven by a wish to provide research of clinical importance to those affected by a cleft lip and palate. Nicola is extremely hard working and ambitious in terms of work quantity and quality. The number of articles she has written and co-authored during the last few years is a convincing example of what she can achieve. I perceive her as reliable, efficient, curious, creative and open-minded, and her ethical standards are high. In addition to high work qualities, Nicola also genuinely cares for people surrounding her, colleagues as well as friends. This combination makes her a wonderful and invaluable colleague.

While we have achieved to publish several papers together during the last two years, we still have many ideas and projects we would like to work on together in the future, if given the opportunity. Nicola is a motivating and stimulating person to work with, and I therefore profoundly hope to have the opportunity to continue my collaboration with her."

Dr Vanessa Hammond

Clinical Psychologist (South Wales) and Chair of the Cleft and Craniofacial Psychology Special Interest Group (SIG)

“The National Special Interest Group (SIG) of Psychologists working in Cleft Lip & Palate teams have worked closely with Nicola Stock and her colleagues over many years. As current chair of the SIG my own involvement has been over the last 6 years. The emphasis over this period has been on working together, both as a full SIG and as a smaller sub-group, to develop a package of psychosocial measures for the Cleft Collective Gene Bank and Cohort Study, including producing a new, cleft specific measure. We have also collaborated over a number of other more clinical issues such as cleft psychology national audit measures and the SIG developed Satisfaction with Appearance measure (SWA).

Working with Nicola has been a positive and productive experience. Nicola has been very supportive of the clinical SIG and we have frequently benefitted from her skills and knowledge. In particular, we are indebted to Nicola for her continuing commitment to including the SIG in the Cleft Collective Cohort Study planning and development. Her diplomacy, honesty and energy have been vital and her ability to balance the sometimes different needs and priorities of research and clinical psychologists working in the field of cleft lip & palate has been hugely beneficial for all parties.”

Dr Angela Shanley

Clinical Psychologist (Oxford) and member of the Cleft and Craniofacial Psychology Special Interest Group (SIG)

“I have known Nicola for 18 months in her capacity as researcher for The Cleft Collective, particularly in relation to her work with the National Psychology SIG for Cleft Lip and Palate, working closely with the members of the SIG to develop the protocols for the Psychology component of the Cleft Collective Research.

From the outset of meeting Nicola she was extremely enthusiastic about the

Psychology component of the CC Research, and in particular recognised the importance of eliciting perspectives from members of the National SIG. Members of the SIG work in different geographical areas, are of different levels of experience and seniority, and are resourced differently according to the cleft service they work in and the way that service is set up.

In order to reach agreement on appropriate Psychological measure, (in particular the cleft-specific questions) Nicola had to draw on the clinical experiences of Psychologists in each of the services, and get a balance between the realities of the Research aims and the need to be inclusive of our views and make use of extensive clinical experience. She worked hard to enable Psychology teams to benefit from the research pragmatically, such as by helping create an overlap between the research questionnaires and the use of these questionnaires for the National Psychology Audit. She worked hard to resolve issues around the practical challenges that the teams might have in delivering a large number of questionnaires as a standard audit and was always responsive to feedback.

I have experienced Nicola as using her research skills in a way which is respectful of the real life research context and responsive to the actual experiences of clinicians and the research participants. She comes across as deeply committed to increasing knowledge and understanding of the psychological issues in Cleft and willing to address the complex day to day issues which can both complicate and also enhance understanding of the actual experience of both clinicians and research participants, our families of children with cleft lip and palate.

Personally I have found Nicola's systematic and responsive approach extremely helpful to me in thinking about the clinical issues I come across in cleft lip and palate, and how complex longitudinal research has potentially a huge amount to offer, despite being challenging to set up in a realistic and meaningful way."

Dr James Kiff

Clinical Psychologist (Cambridge) and member of the Cleft and Craniofacial Psychology Special Interest Group (SIG)

“I am a Chartered Clinical Psychologist within the East of England Cleft Lip & Palate team (Cleft.NET.East) based at Addenbrooke’s Hospital, Cambridge. I am a member of the Cleft Psychology Special Interest Group (SiG) and, more recently a member of the SiG sub-group, supporting Nicola Stock and her colleagues define and identify appropriate psychological constructs and assessment materials and research protocol relevant to cleft patients and their families during core child developmental stages.

Nicola has been instrumental in focusing, coordinating as well as contributing to the combined expertise within the SiG and the sub-group on the key research evidence and issues relevant to the cleft community. She has provided detailed and exhaustive information of current research for the SiG to consider and created a truly collaborative and extensive research program that will provide invaluable data on the impact of having a cleft has on an individual’s psychological well-being and that of their families. Her contribution to date has been invaluable in making the psychological component of the National birth cohort and 5 year-old cohort studies a reality. I have no doubt that the body of research generated though this preliminary work will benefit the patient, their families and clinicians in how cleft services are structured, how psychological practitioners assess, formulate and intervene within services as well as informing the delivery of psychological care.

It is my impression that the thorough examination of the research background, development of research rationale, questions, design and methodology will provide an exemplary foundation for the cleft birth cohort and 5 year-old cohort studies to support and develop clinical understanding and practice, and promote further research in this complex and important area regarding the psychological factors important in cleft, and wider arena of visible/verbal difference.”

Tina Owen

Counsellor/Outreach Specialist for the South West Cleft Team and member of the Cleft and Craniofacial Psychology Special Interest Group (SIG)

“I have known Nicola for approximately 6 years, since she first volunteered at our service as an assistant psychologist. At that time, she was already very well informed about the field of cleft lip and palate and the relevant psychological literature. Her enthusiasm and ability was obvious to all.

Nicola went on to work with the renowned team at the Centre for Appearance Research, but continued her close links with the South West Cleft Team here in Bristol. I have always found her to be diligent and professional in her approach to research. She has an enquiring mind and has never struggled to grasp difficult concepts. She has an ability to grasp the bigger picture while keeping track of the detail. I have also always found her to be compassionate and caring and aware of the person behind the data. When she worked for us in the cleft psychology service as a volunteer she was methodical and capable in all areas. She related very well to the patients and her interpersonal skills were excellent. Her work from this time onwards has gone from strength to strength. She has done an amazing job tracking down and organising the complex research in cleft psychology and brought a unique and important perspective to the field. Her ability to take large amounts of data and distil it into a structured and easily-understood framework is legendary!

It has been a pleasure to work with Nicola on establishing a set of clinically approved research measures for the Cleft Collective. Her ability to meld the research side with the clinical reality is vital to the success of this. Her diplomacy and tact have also been a vital factor. She is someone I look forward to working with and I believe she has great potential to make a further valuable contribution in the field.”

Rosanna Preston and Claire Cunniffe

CEO and Director of Development at the Cleft Lip and Palate Association

“The Cleft Lip and Palate Association (CLAPA) is the only UK charity dedicated to supporting people affected by cleft lip and/or palate (cleft). As a user-led organisation, we work very closely with the CLAPA community to provide a voice for patients and their family.

Nicola Stock began working closely with CLAPA in early 2011 in her role at the Centre for Appearance Research (CAR) at the University of the West England (UWE). CLAPA had secured funding for a pilot project in the East of England and CAR were employed as consultants to provide external evaluation. The aim of the project was to investigate whether having a Regional Coordinator in the East of England would be beneficial for the local cleft community. Funding had only been

secured for 12 months so it was essential that a thorough impartial evaluation was carried out for CLAPA to assess the value of the project and potentially secure additional funding for future years. The project was a success and the Evaluation Report written by Nicola at the end of the project was vital in enabling CLAPA to secure a 4 year grant from the BIG Lottery Fund to employ 3 Regional Coordinators.

The pilot project was the start of what has become an excellent mutually beneficial partnership between CAR and CLAPA, particularly with Nicola. She and her colleagues continue to provide external evaluation for our Regional Coordinators Project and whilst Nicola is careful to maintain the boundary of an external consultant, she is always supportive and enthusiastic about our work and is constantly looking at different and innovative ways to effectively demonstrate the long-term impact our projects can have for people affected by cleft.

Over the last few years, Nicola has led or been involved with a number of studies where we have worked in partnership, most notably her research around the experience of fathers of a child with a cleft and the more recent study on the experience of siblings. The data from Nicola's research is hugely beneficial in helping us to shape our services and in providing an evidence base to help us to generate external funding.

Earlier this year, she played an integral role, along with our Adult Voices Council, in CLAPA deciding on "adults" as the theme for our 2014 Awareness Week. She made available to us her research into the experiences of adults with clefts and worked closely with our Adult Voices Council on the aims and key messages of the week.

A key part of Nicola's role with the Cleft Collective has been to support patient involvement in research. This is a topic that is very important to CLAPA and it has been very beneficial to work with someone who is so positive about patient engagement. Nicola has been involved in recruiting and training patient representatives and consulting with them on patient information leaflets and study design. She has a naturally inclusive and approachable style which makes it easy for patients to contribute and she is clearly genuinely committed to including patients in research. A good example of this is a recent joint presentation at the

Centre for Appearance Research Appearance Matters conference which she gave with two adults with clefts.

We very much look forward to a continued partnership with Nicola and her colleagues at CAR and hugely value her support and the valuable work she undertakes.”

Kerry Humphries

Project Manager of The Cleft Collective Cohort Studies

“My name is Kerry Humphries and I am the project manager for The Cleft Collective Cohort Studies. Nicola Stock works as part of The Cleft Collective team as a Research Fellow. I have worked with Nicola for just over 2 years.

Within those two years, it has been the team’s responsibility to set up and run the project effectively. Nicola has been heavily involved in setting the project up and a valued member of the team. She has been instrumental in making sure that the Patient and Public Involvement (PPI) strand of the project runs smoothly and efficiently. This was an extremely important part of the setting up phase of the project and Nicola has successfully led this aspect from the beginning. It was vital that this work took place in order to gain ethical approval and satisfy the project funders and she worked independently to make this happen.

Another of Nicola’s roles is to lead on the psychological aspect of the study. This involves liaising with the clinical psychologists that are based within each cleft team in the NHS around the UK to pull together a set of psychological measures that can be asked of participants in the study. This work is extremely beneficial to the study but also very challenging. Nicola has persevered throughout this process and as a result has built up a great working relationship with the psychologists. Their input is vital and Nicola has ensured that they have a voice within the project.

The project has funding for a few more years and Nicola’s input would be very much appreciated during this time. Nicola involves herself in all aspects of the study, and is able to see the bigger picture in terms of the project’s short, medium and long term goals. I look forward to continuing working with her.”

Professor Eduardo Keegan

Professor of Clinical Psychology and Psychotherapies, University of Buenos Aires

"I met Nicola Stock in Buenos Aires, Argentina, in the context of an academic trip with the aim of establishing contacts with a number of local research and clinical teams working in areas related to appearance, with the aim of participating in a major European programme. As representatives of the Centre for Appearance Research, Nicola Stock and Martin Persson had previously contacted the leader of one of our research projects, my colleague Professor Guillermina Rutzstein. Professor Rutzstein organized a joint meeting with the people in charge of the Eating Disorders Programme from the National Ministry of Health. The meeting was a success, with government officials expressing interest in cooperating with the future joint project that was discussed that day. Another meeting was arranged with local experts on cleft lip and/or palate from one of our leading university hospitals, with similar success.

On the following day, Nicola Stock and Martin Persson conducted two presentations at the University of Buenos Aires on their current line of work. They explained the problems faced by people affected with cleft lip and/or palate, and described the usual interventions and their results. Also, they explained to our teams the general activities and goals of the Centre for Appearance Research. The presentations were very well received by all members of our research teams.

Since then, we have been in constant contact, as our common project completes all necessary steps for approval and start-up. In my opinion, Nicola Stock is a talented researcher, with a promising line of research. I am certain that Professor Guillermina Rutzstein and all the members of our teams share this view. I will gladly provide any other feedback that might be judged relevant."

Kenny Ardouin

Chief Executive Officer of Cleft New Zealand

"I am writing to express my sincere gratitude to Nicola Stock for her contribution to Cleft New Zealand. After seeing Nicola give such a down to earth presentation

regarding the psychology of appearance at the CLAPA conference in Glasgow in October 2013, I decided to approach Nicola to see if she would be willing to give a similar presentation to our teenagers attending our inaugural national youth camp held in Auckland, New Zealand on December 13-15 2013.

Nicola obliged and went out of her way to deliver an outstanding and very honest presentation via Skype very early on her Friday morning so that it worked in with the timing of our camp programme. For almost all of our teenagers, this was the first time that they had met other people affected by cleft lip and palate, and it was the first time that they were able to discuss the many issues facing an individual affected by cleft with other like-minded people. Nicola's presentation greatly assisted in facilitating these discussions as the teenagers realised that the feelings that they had been experiencing were incredibly common despite being rarely spoken about. Nicola also provided the attendees with many practical pieces of advice to help them overcome difficult situations such as bullying and first impressions. She also provided us with a very helpful handout that people were able to take home with them which contained the key points from her presentation and where to seek further assistance.

Nicola's presentation fitted perfectly with our camp programme, and was appreciated by our camp participants, volunteers and Cleft New Zealand staff as indicated on camp surveys taken after the camp. Her presentation on the Friday night had meant that people felt more open to share stories when we discussed the sensitive topic of bullying on the Saturday.

I am incredibly grateful to Nicola for her support in making our youth camp a success, and for making a tangible difference in the lives of people many thousands of miles away from home. I hope that we can continue to work alongside Nicola in the future, and I hope that she continues with her work which is making a significant difference not only in the UK, but here in New Zealand too."

Katie Stoneman

Research Assistant to Nicola Stock

"I have been working with Nicola as her Research Assistant since September 2013. We have been working on the Regional Coordinators Project, in which we

are externally evaluating the efficacy of Regional Coordinators employed by the Cleft Lip and Palate Association (CLAPA) who aim to improve localised services within their region.

Nicola has been great to work with as she has got the right balance between being supportive yet trusting me to undertake work independently. She has been professional and approachable throughout our working relationship. I have benefitted from working with Nicola through learning research skills but also learning from how she interacts and manages others, including myself. I am happy that I will remain working with Nicola and can continue to learn and develop with her support.”

Participants who volunteered their feedback after taking part in a qualitative study

(Papers 4, 5, 6 and 7)

“I think this is an excellent summary of my experiences and I am very pleased to have been able to help with this.”

“The process and the findings are extremely interesting, relevant and thorough.”

“The results summary made for a fascinating and insightful read that not only reflected my experiences but also demonstrated a great commonality amongst the people that were interviewed.”

“You have produced a fantastic piece of work and I'm sure it will be of a massive benefit to the cleft community and hopefully provide them with some guidance as to where they can improve.”

“I found myself totally relaxed to share my own experiences of being born with a cleft lip and palate and the journey from baby to teenager to adult, despite having not shared this with anyone, even my own family, until the age of 62. It was very liberating and now I want to help as many people who may need the support that I never had.”

“This is an invaluable piece of information that will enhance awareness of clefts among mothers, fathers, family members, parents expecting a baby and those with clefts themselves.”

“This research will stand the test of time and help people recognise the need for psychological care and support. I was delighted that CLAPA chose ‘adults with cleft’ as their theme for this year’s Awareness Week based on Nicola’s research. I wish to give thanks and credit for Nicola’s efforts.”

“You are really hitting the nail on the head with your research - as someone who is approaching the transition into adulthood at the end of treatment and is starting to seriously look to the future, I find myself nodding in agreement when I read through your research. It is great that you are doing the work that you are doing and gaining some traction and getting people’s attention - thank you and keep up the good work!”