Adults' narratives of growing up with a cleft lip and/or palate: Factors associated with psychological adjustment.

Abstract

Background: Growing up with a cleft lip and/or palate presents a number of challenges for those affected and their families. Understanding why some individuals cope well while others struggle is key to psychological research in this field. A better appreciation of the factors and processes which contribute to psychological adjustment to CL/P from the patient perspective would be of value to both researchers and clinicians.

Design: Qualitative data elicited from individual interviews with fifty-two adults born with CL/P.

Results: Inductive thematic analysis identified three main themes: 'background' factors (age, gender, sexual orientation, culture, additional conditions, socioeconomic status and adoption); 'external' factors (treatment autonomy, familial coping and support, salience, public understanding, psychological input and peer support); and 'internal' psychological factors (perceptions of difference, noticeability and teasing, social confidence, internalisation of beauty ideals, valence, expectations of treatment, responding to challenges, social comparisons, acceptance, faith, dispositional style and recognition of strengths and positive growth).

Conclusions: The number and breadth of factors identified in this study is testament to the importance of psychology in the field of CL/P and may offer guidance in relation to developing and assessing the value of psychological interventions. There is a clear role for psychologists in tackling appearance-related concerns, designing materials, supporting patient decision making and improving social interaction, as well as providing specialist psychological support. The findings illustrate the potential degree of individual variation in perspectives and offer insight into the conflicting results found within current literature.

Keywords: cleft lip and palate; psychological adjustment; risk; resilience; adults; intervention

Introduction

Faced with the challenges of a visible difference, the processes involved in treating the condition and the social responses of others, difficulties for individuals with cleft lip and/or palate (CL/P) may arise in relation to low mood, feelings of shame or inadequacy, depression, anxiety, dissatisfaction with appearance, and behavioural consequences, such as social avoidance and aggression (see Thompson and Kent, 2001; Rumsey and Harcourt, 2005; Appearance Research Collaboration, 2009). Such difficulties have the potential to impact upon many areas of an individual's life, including difficulties with social integration and intimacy, educational achievement and work performance, and long-term mental wellbeing (Thompson and Kent, 2001; Rumsey and Harcourt, 2005).

To date, research investigating the psychological impact of CL/P on those affected has produced conflicting results. 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 such as Turner et al., 1998; Hunt et al., 2005; Rumsey and Stock, 2013). 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). Such findings not only imply the use of differing methodological approaches and tools, but also illustrate the complexity of adjustment to a condition such as CL/P (Klassen et al., 2012; Rumsey and Stock, 2013). Furthermore, studies have consistently demonstrated the lack of association between the objective visibility/severity of a condition such as CL/P and psychological adjustment, as well as the relative importance of subjective, 'internal' psychological factors and processes (Moss, 2005; Ong et al., 2007; Brown et al., 2010; Feragen et al., 2010). The question of why some individuals struggle with the challenges of CL/P while others cope well sits at the core of psychological research in this field.

Research in the field of visible difference has begun to identify a number of possible factors which may help or hinder an individual's adjustment. These include 'background' factors, such as age, gender, cultural and religious upbringing and the presence of a condition in addition to the cleft (Thompson and Kent, 2001; Rumsey and Harcourt, 2004; Appearance Research Collaboration, 2009; Feragen and Stock, 2014); 'external'

factors, including availability of social and practical support, relationships with health professionals and awareness of CL/P among the general public (Thompson and Kent, 2001; Rumsey and Harcourt, 2005; Appearance Research Collaboration, 2009); and 'internal' psychological factors, such as sense of self, appearance valence, self-discrepancies, dispositional style and the extent to which an individual relies on the views of others (Thompson and Kent, 2001; Rumsey and Harcourt, 2004; Strauss and Fenson, 2005; Appearance Research Collaboration, 2009).

Despite recent progress, the patient perspective is often lacking. In particular, the views of adults with CL/P are not often heard (Stock et al., in press 2014). Adults' narratives of growing up with a cleft may provide a more holistic reflection of the CL/P journey. In addition, qualitative investigation may contribute understanding of psychological adjustment in a different way to when this question is posed quantitatively or cross-sectionally (Nelson, 2009). Research investigating the effectiveness of psychological interventions in aiding adjustment to CL/P for children, adolescents, adults and families has thus far been limited (Norman et al., in press 2014). An evidence-base to support interventions for those with appearance-related conditions more generally is also lacking (Bessell and Moss, 2007; Jenkinson et al., under review). A better understanding of the role that risk and protective factors and processes play in psychological adjustment to CL/P may provide important knowledge needed to develop and assess new and existing interventions in the field.

Study aim

The aim of this study was to identify possible factors and processes contributing to psychological adjustment to CL/P from the patient perspective.

Method

Design and Participants

In view of the limited understanding available from previous literature, and in order to collect detailed, rich data to inform future research and service provision, an exploratory, qualitative approach was adopted. Qualitative approaches are considered appropriate when the research area under scrutiny is new, or is being explored from a different perspective (Morse and Richards, 2002).

Fifty-two adults with a diagnosis of CL/P participated in this study. Of these participants, 30 were female and 22 were male. Participants were aged between 22 and 77 years, with an average age of 39. Participants had been born with a range of cleft types, including unilateral cleft lip (UCL; n = 4), unilateral cleft lip and palate (UCLP; n = 25), bilateral cleft lip (BCL; n = 2), bilateral cleft lip and palate (BCLP; n = 15) and cleft palate only (CP; n = 6). Participants disclosed a number of additional conditions, including hearing difficulties (n = 19), mental health diagnoses (n = 8), learning difficulties (n = 2), dyslexia (n = 2), heart conditions (n = 2), talipes (club foot; n = 1) and cerebral palsy (n = 1). The majority of participants identified as White British (n = 48), while one participant identified as mixed Caucasian, two participants as British Asian, one participant as White Irish. Most participants were in a long-term relationship (n = 30), and were in full- or part-time employment at the time of interview (n = 47). Twenty-four participants were also parents. Of these parents, eight had children born with CL/P, while 16 had children born without CL/P. Parents of children with CL/P had an average of two children, with one child affected by cleft. Parents of children without CL/P also had an average of two children. None of the participants in this study identified themselves or their children as having an associated syndrome.

Participants were recruited from across the UK, with representation from all UK regions. Fifty percent of participants were recruited through the UK-based charity, the Cleft Lip and Palate Association (CLAPA), while the remaining fifty percent were recruited through other means, including a UK-wide university press release which was picked up by numerous national newspapers, charities and websites, and through the use of appropriate social media tools.

Once informed consent had been obtained, individual, free-response recorded telephone interviews were conducted until saturation was achieved (fifty-two adults). In qualitative research, saturation occurs when no new information is being obtained, thus determining the sample size (see Braun and Clarke, 2006). In a qualitative study such as this, it is the content of the interviews, rather than the number of participants, that is considered important (see Howitt and Cramer, 2011). All interviews were conducted by the first author; this researcher has a background in psychology research and is trained in qualitative interviewing techniques. Although the interviewer aimed to cover a number of key topics within the course of each interview and had prepared prompts to elicit further information when necessary, participants were otherwise allowed the freedom to relay their own story. Topics which the interviewer aimed to cover included: family history of cleft; early

childhood memories; school experiences; the treatment journey; social experiences; transition into adulthood; decision-making around starting their own family; experiences of having children (where applicable); past and current support needs; and reflection of their journey as a whole. Interviews lasted 45 minutes on average.

Centralisation of cleft care in the UK

Following a report commissioned by the Clinical Standards Advisory Group (CSAG) in 1998, cleft care was significantly reorganised in the UK (Sandy et al., 1998). Prior to this reorganisation of care, no standardised treatment protocols were implemented across teams and psychology was not offered as part of the service. It is thus important to keep in mind that all of the adults who took part in the present study had received treatment which preceded service centralisation. Nonetheless, it is hoped that the findings of this study will be applicable to those children who are currently engaged in the revised treatment pathway, as well as to those adults who have already been discharged from the cleft service.

Ethical considerations

Ethical approval was obtained from the University of the West of England Research Ethics Committee. The research adhered to the British Psychological Society's Code of Ethics and Conduct (BPS, 2009) at all times. Prior to taking part in the study, participants were made aware of key issues including confidentiality and their right to withdraw. In addition, participants were advised that the researcher was not able to provide them with advice or counselling at any point; participants were directed to relevant organisations should they wish to seek further information and/or support.

Data analysis

Thematic analysis (TA) was used to analyse the interview data. TA is primarily a method for identifying and organising patterns within a rich data set, though it can also be used to interpret various aspects of the subject matter (see Howitt and Cramer, 2011). For this study, an inductive, data-driven approach to analysis was taken, adopting a pragmatic framework (see Fishman, 1999). The goal was to report a rich, overall description of the whole data set, rather than explore particular aspects in more detail. In accordance with Braun and Clarke's guidelines (2006), the following steps were taken:

1) Becoming familiar with the data

2) Identifying interesting features of the data

3) Searching for themes

4) Reviewing themes

5) Defining and naming themes

6) Producing the report

Analysis was seen as a recursive process, and detailed notes were written throughout. Themes were subsequently chosen for their prevalence and/or their importance (or 'keyness') in relation to the research question. Emerging themes were initially identified by the first author. Themes were then checked and discussed until agreement was reached between all three authors. A summary of the resultant themes was also sent to participants to confirm the accuracy of the analysis, and their feedback was incorporated into the final report (*see* Braun and Clarke's 2006 guidance, Step 4; Yardley, 2000).

Results

The factors identified through thematic analysis were organised into three main themes consistent with previous literature: '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). Each theme is described below with exemplar quotes and summarised in Table 1. Pseudonyms have been used in order to preserve participant anonymity.

THEME 1: 'Background' factors

Age

Although many participants believed cleft was a lifelong condition, some felt they had become more accepting of their cleft over time.

I think I started becoming aware of it and thinking 'ah I wish I didn't have this'…but then as you grow up and get on with life you realise that it's nothing to hold you back at all – Sean, 47, BCLP

Other things become so much more important as you get older, and you sort of think 'well what was I worrying about really?' - Lauren, 58, UCLP

Gender

A number of participants commented on possible gender differences in relation to appearance.

I think because I'm a guy and I play a lot of sport, people just assume I broke my nose playing rugby or something… Maybe it's different for girls, I don't know…there's a lot of emphasis on having a pretty face – Mark, 27, UCLP

I was quite thin growing up, very skinny and underweight, like a 'weakling'…being male I don't think that helped my case, so I was conscious of that too – Robert, 60, BCLP

Sexual orientation

In a similar way, two participants felt there was a lot of emphasis on appearance among the gay community.

I would think it's fair to say that in the gay world, people are much more image conscious, it's much more of a fickle dating scene, so it perhaps has been a disadvantage for me in that area – Jason, 37, BCLP

Ethnicity, culture and religion

For the few participants who identified themselves as being from minority ethnic backgrounds, cultural and/or religious beliefs had played a significant role in their experience of CL/P.

Being Asian, the real issue even when I came into my thirties was marriage, because arranged marriages are very much about looking the part…all they could see when they looked at me was my scars…as far as they were concerned I didn't look right for their daughters, and the fact that I'd gone to Oxford University and had come from a good family and I was a nice person was irrelevant – Samuel, 44, BCLP

Additional conditions

A small number of participants who took part in this study disclosed themselves as having a condition in addition to the cleft. The most common conditions included learning disabilities and difficulties associated with hearing. Participants felt that having an additional condition had impacted on their social experiences and education, and believed this impact to be greater than having a cleft alone.

For me, the cleft was never that much of an issue. It was the learning disability that had an impact on my daily life – Richard, 26, BCLP

My hearing fluctuated quite a bit…sometimes I would mishear something and then feel stupid, because I didn't get what people were laughing at… I became quite anxious that I was missing out on things. It also wasn't very cool, because I would sit near the front of the class to hear the teacher and everyone probably thought 'oh, what a [geek]' – Hannah, 50, CPO

Socioeconomic status

Some participants commented that their childhood environment had an impact on their adjustment to CL/P.

We didn't have a lot of money when I was younger…it cost a lot to ride the buses back and forth to appointments, so I think I missed quite a few – Natasha, 26, UCLP

Adoption

Two participants in this study had been adopted when they were young.

My mum couldn't cope with me having the cleft lip and palate basically and that's the reason I got adopted into a nice family that could deal with the cleft. My adoptive mum used to be a nurse with children so she had a lot of experience – Emma, 28, BCLP

THEME 2: 'External' factors

Treatment autonomy

Many participants recalled 'traumatic' experiences of hospitals during childhood. This was linked to negative interactions with Health Professionals and a lack of autonomy in regard to treatment.

When I was a child they talked amongst themselves in their own language and I didn't really know what was going on or what plans were being made for me…and I said yes to everything because I wanted to look better and I wanted to feel better…it was very disempowering - Grace, 28, UCLP

My surgeon wanted me to have more done but I said 'no, I really can't do this' and he obviously wasn't impressed… He told me 'you don't look right', and I was doing a science degree and he said 'if you want to work at a higher level you will need to get your face [fixed]' – Spencer, 46, UCLP

Family appraisals and coping

For some participants, the perception that their parents hadn't coped well with their cleft and its treatment had a lasting influence on their own adjustment.

My dad wasn't around much, he just couldn't deal with it…my mum always found it hard to accept…she saw it as a disability…she thought it was a punishment for something…there was no support to help her get past it – Nina, 37, BCLP

Familial support

Similarly, some participants had never spoken with their parents about their cleft, while others felt they had been well supported within the family.

To a great extent it was just brushed under the carpet and never dealt with properly…it was a bit of a taboo subject really – Chloe, 37, BCLP

My family's been with me all the way, they tried to answer any questions I had and showed me photos from when I was born…I had a good understanding of what happened, so I've got no problem talking about it – William, 22, UCL

Salience of the cleft

Many participants felt that situational factors had an influence.

It all depends on where you are, who you're with…sometimes you hardly think about [cleft] and then someone will say something or something will happen and…you're back to square one – Lucy, 36, BCL

<u>Public understanding of CL/P</u>

Participants also spoke about the lack of understanding of cleft among the general population.

People stare quite often and have a lot of questions…education and awareness generally; there doesn't seem to be a lot of knowledge out there about what [cleft] actually is…and what the impact is and that it's not the end of the world – Kate, 35, UCLP

There wasn't much awareness in my school…my teachers didn't understand, or know how to help – Heather, 49, UCLP

Some people seem to think it only happens in India, or in places where people live in poverty, because of those adverts that you see on TV – Liam, 54, BCLP

<u>Degree of psychological input</u>

The majority of adults in this study had not been offered any professional psychological support throughout their childhood. Many felt this would have been helpful.

Some level of counselling, psychological support…someone just to be very honest with…in my teens especially I definitely would have benefitted from that – Jason, 37, BCLP

<u>Peer support</u>

Many adults in this study had not met someone else affected by CL/P while growing up. Although not all participants said they wanted to engage in peer support, many discussed the potential benefits of meeting others with cleft.

It's just about realising for the first time that you're not the only person in the world with a cleft… Just seeing somebody else and talking to somebody else who has been through similar experiences to you – Leah, 35, BCL

CLAPA didn't exist when I was younger, the internet didn't exist…so there wasn't really that external support network other than the family…an outside point of reference would have been useful – Jacob, 37, UCLP

THEME 3: 'Internal' psychological factors

A wide range of factors were identified for this theme, highlighting the multifaceted nature of adjustment and the wide range of experiences reported within this study. Each of these factors are introduced below, followed by examples of each differing viewpoint identified within the data.

Perceptions of 'difference'

While some participants felt that having a visible difference gave them some individuality, others perceived this difference to be very negative. Some did not perceive themselves to be different.

I like to stand out slightly I think, it's nice to have some individuality and be unique – Sean, 47, BCLP

As I got older I became more aware that I was different. I stood out for all the wrong reasons and I hated it – Grace, 28, UCLP

To be honest I don't see myself as being any different from anyone else – Lucy, 36, BCL

Perceptions of noticeability

Some participants felt their 'difference' was very obvious, while others didn't feel their cleft was noticeable to others. This could be viewed in either a positive or negative light.

I've seen other people's clefts, and theirs is actually really noticeable, whereas I think mine's quite good…my speech isn't that bad, I've had it really easy to be honest…I think most people don't even realise I have it – William, 22, UCL

I think because it's very visible, it makes you confront it, if it's not hidden away then you've got to confront it on a daily basis – Ben, 35, BCLP

Perceptions of teasing/bullying

Almost all participants reported being teased about their cleft at some point. However, participants' perceptions of this varied.

I don't think I was teased any more than anyone else…I think there are kids who take offense to things that are said, but for me…it just went over my head, I didn't really take it to heart – Natasha, 26, UCLP

School wasn't easy for me...but I had my close knit people of about six friends and that was it...I hung around with them, and tried not to let it bother me – Emma, 28, BCLP

Social confidence

Participants commented that being socially competent was important in overcoming social barriers.

I never had much luck with girls but I think that was me being shy and introverted more than anything...not really knowing what to say and generally being quite awkward – Ben, 35, BCLP

I've always been confident and assertive and made friends very easily, and I'm quite an approachable person and good at listening...so I think my confidence overcame any problems with my speech or the way I looked – Olivia, 47, CPO

Internalisation of beauty ideals

While some participants seemed quite susceptible to societal ideals and media messages about beauty, others believed that growing up with a cleft had helped them to recognise and challenge the 'beauty myths' from an early age.

We live in a world which is obsessed with appearance and youth and beauty, so when you don't conform to a certain standard it can be very difficult – Poppy, 29, BCLP

Everyone's got something they're not keen on don't they? For me, it was just a bit of a misshapen nose – Eric, 32, UCLP

There are friends I recall from school...they had a regular pretty face and they don't have the same confidence that I do, and they don't have the same ability to talk to people...beauty only gets you so far doesn't it? – Olivia, 47, CP

Appearance valence

For some participants, appearance was an important part of their self-esteem. This seemed to be associated with the belief that surgery was a quick way of improving other aspects of life.

I think the mismatch came when I felt I wasn't pretty… I believed that who I was hinged a lot on the way I looked – Hannah, 50, CP

Growing up I was always looking forward to the next operation; I still see it as a way to solve things… even now I can't go out of the house without make-up on and it's a complete mask really – Grace, 28, UCLP

Expectations of treatment

Participants discussed the importance of psychological input in relation to managing patients' expectations of surgical outcomes.

When I had the operation I had this silly idea in my head that I would be given a book of pretty faces…and that I could choose which one I was going to look like… I thought [the operation] would change my life, I thought I would be 'normal'…no one gave me a realistic picture – Louise, 45, UCLP

Strengths versus deficits

Participants also believed that being involved in a variety of activities, and focusing on their strengths, had contributed to their adjustment.

I played quite a lot of sport when I was younger…being part of a team really helped because everyone is equal, and I think you get quite a lot of respect… I got involved in a wide variety of activities and focused on what I was good at… I think that stood me in good stead – Ben, 35, BCLP

Responding to challenges

While reflecting on their journey as a whole, many participants believed that having a cleft had presented them with a number of challenges throughout life, and the way in which they had responded to these challenges had shaped their development.

There were occasions when I was young where I could have sunk into a corner…and you have to make a decision about how to respond…I suppose I've seen it more as a challenge…it enabled me to dig my heels in and think 'I'm not going to let this ruin my life' – Oscar, 77, UCLP

Social comparisons

A common statement made by participants was that in comparison to some others, they felt 'fortunate'.

Other people go through much more horrendous things than I did and still come through on the other side – Nina, 37, BCLP

Acceptance

Many participants were accepting of their cleft and viewed it as being a part of who they are.

I wouldn't go so far as to say I'm glad I was born with a cleft, but it has helped define who I am and I am part and parcel of that person with a scar on her face – Kate, 35, UCLP

Faith and spirituality

A few participants discussed how their faith had helped to give them a sense of 'identity' and 'belonging'.

My wife and I are Christians…we have a very strong sense of identity…so even though I was naturally nervous, shy, lacking in self-confidence…my self-esteem has been transformed by my faith – Anthony, 48, UCLP

Dispositional style

Having a positive outlook on life and a good sense of humour were identified as important factors by many participants.

You have to find the positives…and have a sense of humour about it I suppose…I could get bogged down in thinking 'oh poor me', but where would that get me? – Lauren, 58, UCLP

Recognising positive growth

Participants were able to identify a range of positive consequences of growing up with CL/P.

It's made me a better person. I'm a lot more understanding, I wouldn't judge anybody...I've met some lovely people and I've done things that I probably wouldn't have done…I wouldn't change it for anything – Abigail, 33, BCLP

Discussion

This study has identified a number of potential factors which may help or hinder adjustment to CL/P, according to the perspectives of adults who have grown up with the condition. These factors will now be discussed in the context of previous studies.

'Background' factors

Age

Participants in the present study felt that their outlook with regard to their cleft had improved over time. For many, accepting the cleft was part of a natural process of 'growing up' and gaining perspective on the relative importance of CL/P in relation to other personal qualities and aspects of life. However, CL/P remained a consideration for older participants, and individual variation in adjustment among participants was substantial at all life stages. These findings are consistent with the limited number of other studies in this area (Appearance Research Collaboration, 2009; Hamlet and Harcourt, in press) and may explain why little research has found conclusive results in relation to age. In addition, although adjustment to a visible difference is generally characterised as being relatively stable, adjustment has been shown to fluctuate within individuals over time, and in relation to different situations and life events (Appearance Research Collaboration, 2009). The findings from the present study therefore imply that it may be helpful to identify potential points of risk or growth according to life stages and events, rather than assume age to be a direct linear factor in adjustment to CL/P.

Gender

Previous research has also attempted to understand the role of gender in adjustment to CL/P. Within the present study, gender was predominantly discussed in relation to appearance. Some participants felt that having a cleft may be worse for girls, due to a societal emphasis on petite, feminine facial features. A small number of studies have found appearance-related self-esteem and dissatisfaction with appearance to be higher in girls with CL/P than boys, and to be mediated by greater perceptions of peer harassment (Leonard et al., 1991; Feragen and Borge, 2010). In the general population however, appearance concerns have also been found to be prevalent in boys, particularly in relation to the drive for muscularity (Pope et al., 2000; Grogan and Richards, 2002; Grogan, 2007). In the present study, a number of male participants described feeling 'weak', 'skinny'

and 'underweight', especially during their adolescent years. Most men in resource-rich countries report a preference for being tall and muscular (Tiggemann et al., 2008); thus for males born with a cleft who may find their physical development to be behind that of their peers (Persson et al., 2007), this may be particularly challenging. The current findings thus point to some gender-specific concerns in relation to appearance.

Sexual orientation

In the present study, two male participants identified as being homosexual. These participants discussed having CL/P as being a potential disadvantage, due to their perception of the emphasis placed on appearance within the homosexual community. To the authors' knowledge, issues of sexuality have not been previously discussed in the area of CL/P. More generally however, research has found homosexual men to report feeling more pressure to conform to appearance ideals, spending more time discussing appearance with others, and experiencing more appearance dissatisfaction than heterosexual men (Morrison et al., 2004; McArdle and Hill, 2009; Jankowski et al., 2014). The present study suggests that sexual orientation could be a topic which requires further consideration within the general appearance field, and may also be indicative of a potentially vulnerable subgroup in relation to visible difference and/or CL/P.

Ethnicity, culture and religion

Only two 'British Asian' participants and one 'Mixed Caucasian' participant identified as being from a different ethnic/cultural group to the rest of the sample; yet the influence of cultural and religious expectations and beliefs on adjustment was evident for these few. Research has previously demonstrated the authority of such beliefs over community attitudes toward visible difference and on those who are affected (Appearance Research Collaboration, 2009; Hughes et al., 2009). In the present study, participants discussed experiencing difficulties in being accepted by others in their community. In adulthood, having a cleft was especially problematic in tackling stigma and in finding a marriage partner. Although this study raises some important points, larger samples are needed to understand the impact of CL/P on different cultural groups, and of the additional and/or alternative support which may need to be implemented in response to culturally-specific support needs.

Additional conditions

A high prevalence of additional conditions, including learning difficulties, dyslexia, autism, AD/HD and developmental delay has been identified among children with CL/P (Feragen and Stock, 2014). In addition, hearing difficulties are known to affect many children born with a cleft (Sharma and Nanda, 2009). In the present study, those who reported having one or more additional conditions described their belief that such conditions had impacted to a much greater extent on their education, employability and social integration than the cleft itself. This suggestion is in line with recent research which demonstrated that when additional conditions are accounted for, children with a cleft alone report scores of psychological adjustment which are comparable to the general population, while a high percentage of the difficulties reported lie with those children with one or more conditions in addition to the cleft (Feragen et al., 2014). Despite clear evidence that the presence of additional conditions can affect psychological adjustment (Feragen and Stock, 2014), relatively little is known about this group of patients and how to support them. The current findings therefore pose the identification of children who may be at risk of increased psychological difficulties to be an important future consideration.

Socioeconomic status

While little work in the field of CL/P has investigated the specific challenges of those from lower income households, one study suggested that socioeconomic factors, including lower household income, poorer overall health and lower educational status was associated with poor maternal mental wellbeing in mothers of children with CL/P (Dabit et al., 2014), a finding which also relates to the general population (e.g. Turney, 2011). The impact of socioeconomic factors was also alluded to in the current study by a minority of participants. Given that the incidence of CL/P is thought to be higher among low income communities (Yang et al., 2007), the current findings suggest that future work may need to account for these differences when necessary, and to find ways of improving access to treatment and support for this potentially vulnerable population.

Adoption

Although only two participants disclosed their adopted status in this study, both participants attributed their adoption to being born with CL/P and their biological parents' subsequent inability to cope. This could be an important area of CL/P which is under-researched. For all parents with a baby born with CL/P, parental

perceptions about the condition and the quality of information they receive early on have lasting effects on their psychological status (Vanz and Ribeiro, 2011). The present findings are in line with one recent study (Hansson et al., 2013) and together suggest that a number of factors, including adoptive parents' decision to adopt a child with CL/P, their experiences of this process and the subsequent adjustment of the adopted child require further investigation.

'External' factors

Treatment autonomy

Several studies have identified a discrepancy between the views of health professionals (HPs) and their patients, particularly in relation to treatment choices in CL/P (Knapke et al., 2010; Nelson et al., 2012a). In the current study, this discrepancy was reported in relation to participants' childhood experiences and, despite improvements in the way cleft services are delivered, also in relation to more recent experiences in adulthood. These findings are in concordance with previous research suggesting that the relationship between the HP and the patient is of vital importance to the patient experience and to the treatment outcome (Nelson et al., 2012a). Equally, research has demonstrated that the patient's subjective satisfaction with the outcome of their treatment is likely to be of more significance to their subsequent adjustment than objective measures of treatment success (Ong et al., 2009; Cadogan and Bennum, 2011). Over the lifespan, a patient and/or their family are faced with a range of bewildering and successive options regarding their/their child's treatment (Rumsey and Harcourt, 2005). Patients who are emotionally distressed may have a reduced capacity to assimilate information and may therefore perceive a lack of support regardless of the HPs' actual competence and knowledge. The findings of the current study imply that HPs require an awareness of patients' support and information needs, as well as good communication skills, in order to support patients to feel empowered in making informed and realistic treatment decisions. Involving a psychologist in this process may also help to alleviate psychological distress and aid communication between HPs and patients.

Family appraisals and coping

A number of participants in the current study reported family members to express negative appraisals of CL/P and to struggle to cope with the cleft and its treatment. While the birth of a child is an emotive time for any

family, having a child with a health condition such as CL/P poses additional challenges (Nelson et al., 2012b). The manner in which the individual's family perceives the cleft and its treatment, as well as the family's ability to cope with these implications may be an important factor in the child's own adjustment (Baker et al., 2009; Nelson et al., 2009; Berger and Dalton, 2011; Nelson et al., 2012b), as was indicated in the current study. Much research has discussed the significance of making appropriate support and information accessible to families throughout their child's developmental trajectory (see Collett and Speltz, 2007; Nelson et al., 2012a; Rumsey and Stock, 2013). Prior to entering into social interaction with their peers, the child's main reference is the family environment, and the family continues to have a large influence throughout the individual's development (Nelson et al., 2012b). The findings of this study support the suggestion that identifying families who may be at risk of psychological difficulties and offering them appropriate support may also create opportunities for better psychological adjustment in the child.

Familial support

Correspondingly, the level of support which the family is able to offer their child to cope with the impact of the cleft is likely to affect the child's adjustment (Nelson et al., 2012b). The findings from the present study suggest that when a family is open with their child in talking about the condition and any concerns the child has, the child in turn is better able to speak openly with their peers and to deflect negative comments. Some participants in the current study described being made to feel 'unique' or 'special' by their family because of their cleft, while others reported being treated 'normally' or 'the same' as everyone else. While these reports seemed to be associated with better adjustment, several participants reported that the cleft had 'never' been talked about within the family, that it was a 'taboo' subject and they had been expected 'to get on with it'. A small number of participants had also felt they could not speak about their cleft for fear of upsetting their parents. It may be that some parents feel ill-equipped or reluctant to speak to their child about CL/P (Stock and Rumsey, in press 2013), which in turn affects the family as a whole. The issue of familial support and how to improve familial relationships is a complex area for research to address, since it is partially built on interpretations and attitudes which are especially difficult to capture.

Salience of the cleft

For many participants in this study, CL/P was expressed as an underlying factor which became more or less salient depending on situational factors. Participants described feeling more anxious in situations where the cleft may be perceived as more prominent, for example, meeting new people at a social event or attending a job interview. Similarly, a single negative social interaction was reported to bring cleft-related concerns to the surface. Previous research in the area of visible difference has linked increased levels of salience to higher levels of social anxiety and social avoidance (Appearance Research Collaboration, 2009; Moss et al., 2014). Current findings suggest that it may be helpful to equip individuals with CL/P with cognitive tools and social strategies to cope with these concerns as and when they arise, as well as to focus on ways of tackling specific events which may provoke anxiety.

Public understanding of CL/P

While offering individuals with CL/P strategies to cope with difficult situations is undoubtedly useful, a much larger challenge is to directly tackle the stigma and discrimination which is still being identified within society today (Grandfield et al., 2005; Changing Faces, 2008). Participants in the present study perceived a lack of awareness and understanding of CL/P among the general population, which also extended to those playing a key role in their care and upbringing, including General Practitioners (GPs), dentists and teachers. A number of UK-based charities have made great steps in raising awareness of visible difference (see Changing Faces (www.changingfaces.org.uk), the Healing Foundation (www.thehealingfoundation.org.uk) and the Cleft Lip and Palate Association (www.clapa.com). Conversely, some campaigns may detract from this. For example, participants mentioned that fundraising advertisements for children affected by CL/P in resource-poor countries contributed to the public's belief that CL/P does not occur within Western society. Findings therefore imply that additional work to raise awareness of the challenges of visible difference and to promote diversity in appearance is needed. In particular, participants in the current study strongly expressed the desire to communicate that being born with CL/P does not have to be socially or educationally disabling if suitable support and opportunities are provided.

Degree of psychological input

The majority of participants in this study had not received psychological support while growing up. Some believed they had not needed professional input, while others felt this would have been a useful outlet, particularly in the absence of perceived social support from family or peers. Previous work in the area of visible difference has proposed a stepped model of psychological intervention for this (and other) population(s) (see Clarke et al., 2013). This approach suggests that all healthcare providers have a vital role to play in promoting positive psychological adjustment, based on a range of interventions of varying type and intensity of need. The findings of the present study are in line with the previous suggestion that a preventative developmental approach to care may help to reduce the risk of psychological distress in adulthood (Stock et al., in press 2014). Access to psychological support for those who have already left the service also requires additional consideration (Stock et al., in press 2014).

Peer support

A number of studies have discussed the potential benefits of meeting someone else who has been affected by CL/P, particularly if the other person is slightly further ahead in the treatment pathway (Nelson et al., 2012b). This level of support affords individuals opportunities to share stories with someone who has similar experiences, and profit from mutual social support. In the present study, many participants had not had this opportunity when growing up, and a further number of participants had never met anyone else with a cleft (even up to the age of 77 years). The findings indicated that a point of information and support outside of the family unit is valuable during childhood, adolescence and adulthood. The Cleft Lip and Palate Association (CLAPA) now offer peer support to anyone affected by cleft, along with standardised training for those volunteers who provide it. This not only allows people affected by CL/P to receive a high standard of information and support from those with personal experience, but also offers them an opportunity to be advocates for others. Peer support groups for parents have already shown promise (Douglas, 2012) and may thus offer another practical application of these findings.

'Internal' psychological factors

Perceptions of 'difference'

During early development children start to become more self-aware, and are increasingly aware of themselves in relation to others. Many of the participants in this study reported feeling 'different' from their peers from the age of seven to eight years, which also seemed to be a key age for assimilating negative messages from perceived teasing and bullying. For others, primary school had been an enjoyable experience, but the transition to secondary school and their adolescent years had been more difficult. During adolescence, 'blending in' and feeling included in a social circle are key facets for psychological wellbeing (Liossi, 2003). As the result of appearance concerns, speech, hearing and communication issues, and/or associated difficulties, individuals with CL/P may thus feel vulnerable in comparison to their peers (Rumsey and Harcourt, 2004). In contrast to these ideas, some participants in the current study reported feeling content that they were different from the norm and placed high value on individuality and diversity. Others did not believe they were different, or believed that everyone was different from one another in some way, and thus their condition did not make them stand out any more than anyone else. These beliefs seemed to be related to less psychological distress, while those who felt they were different in a negative context, or believed themselves to be somehow 'flawed' reported more negative experiences in relation to the cleft. The findings of this study demonstrate that perceptions of 'difference' may be key in understanding how individuals with CL/P process and interpret social experiences and life events. These perceptions and interpretations may mediate the social experiences that researchers and clinicians attempt to measure, and therefore need to be better understood in order to support individuals with CL/P who may be struggling with social interaction.

Perceptions of noticeability

Linked to 'perceptions of difference' was the individual's perception of how noticeable their condition is to other people. Multiple studies have now demonstrated that the objective visibility or the severity of a condition does not predict adjustment (Moss, 2005; Ong et al., 2007; Brown et al., 2010; Feragen et al., 2010), indicating the relative importance of the individual's subjective perceptions and psychological processes. In the present study, many participants did not believe their cleft to be noticeable to others. Some attributed this to having received a high standard of treatment, while others did not feel their cleft had been 'as severe' to begin with. Although many participants felt their cleft was visible to others, some believed this had helped them to confront any fears they had because there was 'no hiding from it'; a suggestion also posed by previous literature

(Lansdown, 1997). In contrast, the perception that their condition was highly noticeable to others and the notion of this being a significant disadvantage seemed to be associated with more negative self-perceptions and social experiences. Findings imply that ways of fostering more positive self-perceptions need to be identified and encouraged (also see Clarke et al., 2013).

Perceptions of teasing/bullying

Teasing and/or bullying is reported by approximately half of all children and young people in the United Kingdom and United States (Chamberlain et al., 2010; Bradshaw et al., 2011). Although children with additional difficulties such as an altered appearance, communication issues and potential developmental difficulties have often been assumed to be a particular target for teasing/bullying, research suggests that this is not always the case (Carroll and Shute, 2005). In the present study, approximately half of participants believed themselves to have been badly teased or bullied, while the remaining participants had not perceived any teasing/bullying, or had perceived the teasing/bullying they had experienced to be 'normal' or 'negligible'. While this discrepancy is interesting in itself, a further proportion of participants who reported being teased/bullied did not feel this experience had a deep or lasting effect on their social or emotional development. Again, this finding may be linked to the way the individual interprets social situations, as well as to their available resources and their perceptions of their ability to cope. Additionally, a number of protective factors may play a role. Research has suggested that friendships may act as a protective factor from depressive symptoms and negative self-perceptions of appearance (Feragen et al., 2010). In the present study, some participants commented that they had a small but close circle of friends who were accepting of their condition, and thus comments from people outside of this social circle mattered less. Those who are vulnerable to teasing are more likely to incorporate these experiences into their own self-perceptions (Feragen et al., 2010). Thus, the ability to reject teasing/bullying may also be indicative of a degree of emotional resilience (Feragen et al., 2010). The present study suggests that perceptions of social interactions appear to be key in relation to psychological adjustment to CL/P, and should thus form an important part of any psychological intervention. Participants also commented that anti-bullying policies (see for example Department for Education, UK, 2014) had not been widely implemented in schools when they were growing up. In relation to visible difference more

specifically, it has been suggested that training and supporting teachers in relation to the issues pertaining to these conditions may help to facilitate adjustment in children (Frances, 2003).

Social confidence

Early research in this area found that individuals with a visible difference were viewed more positively than those without a facial difference if they possessed good social skills (Rumsey and Bull, 1986; Partridge, 1990). In the present study, a reported lack of confidence in social situations was associated with the perception of less positive social experiences, while some participants believed that a higher level of social confidence had helped them to overcome the difficulties associated with looking and/or sounding different. Although sociocultural messages also need to be challenged directly, this study also supports the view that given the right tools (see for example Kapp-Simon, 1995; Kleve et al., 2002), it can be possible for the individual to feel more confident in social situations and, as a consequence, to elicit more positive social responses from others.

Internalisation of beauty ideals

With extremely powerful industries advocating the importance of beauty, such messages can be difficult to ignore. In the present study, some participants found the pressure to conform to societal 'ideals' very challenging. In contrast, others felt that having been born with a condition which was visible to others had helped them to unpick the beauty 'myths' early on in life. Some participants believed they possessed a higher level of confidence, self-awareness and self-esteem than some of their friends who were not born with a visible difference, which they attributed to their experiences of CL/P. Participants also commented that dissatisfaction with appearance was widespread, irrespective of whether a person had a visible difference or not. Thus, a 'misshapen nose' or a 'facial scar' was perceived by many participants as one such minor burden. While appearance dissatisfaction has been termed 'normative' to demonstrate the extent of appearance concerns, mere exposure to societal pressures is not considered enough to result in body image disturbance (Thompson et al., 1999). The present study supports the idea that the degree to which an individual internalises sociocultural ideals could be one intervening factor (see Dittmar, 2005). Although further research is needed to explore the characteristics of those individuals who are successfully able to 'bust' the beauty myths (Thompson et al., 1999), learning to challenge the sociocultural messages which surround us all, particularly during childhood

and adolescence, may be a powerful preventative strategy for psychological intervention, both in general (Yager et al., 2013) and in the field of CL/P.

Appearance valence

Appearance valence refers to the extent to which appearance is important to self-esteem (Rumsey and Harcourt, 2005; pp. 53). In the present study, it was clear that some participants placed a high value on appearance, particularly in comparison to the value placed on other personal characteristics. For these participants, CL/P seemed to be much more of a salient and ongoing concern than for those participants espousing low appearance valence. Participants with high appearance valence also seemed to report more favourable attitudes towards the possibility of additional surgical interventions, and some viewed surgery as a quick way of improving other aspects of life in the long-term. This finding relates to research in the wider field which suggests that individuals with a higher level of appearance valence may be more prone to interpreting and recalling appearance-related information from the social environment as negative (Moss and Carr, 2004), perceive a greater discrepancy between the way they think they look and the way they think they should look, and subsequently experience increased levels of social anxiety due to a fear of negative evaluation by their peers (Thompson and Kent, 2001). The findings of the present study suggest the importance of psychologists in challenging the perceived importance of appearance, to help patients to reduce the amount of time and attention they pay to appearance and to support the individual to identify other positive qualities.

Expectations of treatment

Within the present study, several participants had considered having additional surgery, or were looking to surgical intervention to 'solve' difficulties related to psychological wellbeing and social function. While biomedical interventions are useful for some, an unrealistic view of the impact surgery could have on self-esteem and on external parameters (e.g. establishing new relationships or getting a new job) is associated with poorer psychosocial outcomes (Sarwer et al., 1998; Honigman et al., 2004). Some participants in this study commented that in hindsight, their expectations of surgical results had been unrealistically high. Advertisements for cosmetic surgery often encompass a degree of glamour and sensationalism, and general interest in cosmetic surgery as a way to 'improve' appearance and/or psychosocial wellbeing is increasing

among the general population (Frederick et al., 2007). In addition, sensitive or emotional decisions are rarely made in line with theories of decision making which assume rationality (Rumsey and Harcourt, 2005, pp. 173). In order to avoid the unnecessary uptake of surgical interventions and to prevent disappointment and distress arising from unrealistic expectations of surgical outcomes, the present study suggests that psychologists have a role to play in helping the prospective patient to clarify priorities, attitudes, knowledge, motivations and confidence, in order to support decision making (Rumsey and Harcourt, 2005, pp. 173; Clarke et al., 2013).

Strengths versus deficits

Many participants in this study reported that their adjustment to CL/P has been facilitated by involvement in a variety of social activities and interests. Coherently, and in parallel with findings related to appearance valence, these participants considered their self-esteem to be based on many characteristics and attributes aside from their appearance. Participants commented that being involved in activities both inside and outside of school had linked them to a wider social circle and had allowed them to focus on their strengths rather than any perceived deficits. In particular, several participants felt that being involved in sport allowed them to become part of a team and generate respect from their peers. Research in the wider field has shown that placing high value on positive characteristics and qualities can facilitate adjustment in those living with a visible difference (Strauss and Fenson, 2005; Egan et al., 2011). More generally, success in the academic or extra-curricular sphere may support the development of self-efficacy and help to create a sense of belonging to a valued social group (Gilligan, 2000). Research in the field of eating disorders has also suggested that participation in sports may help individuals to value their body for its function, rather than its appearance (see Hausenblas et al., 2008). The findings of the present study support the call for more of a focus on patients' strengths, to balance out the emphasis to date on problems and shortfalls (Strauss, 2001; Nelson et al., 2012b; Rumsey and Stock, 2013). Supporting patients to explore their interests, as well as helping individuals and families to identify and foster their strengths, could be useful tools for future psychological intervention.

Responding to challenges

Closely linked to the concepts of coping and resilience is the ability to respond positively to, and grow from, challenges (Luthar, 2006). In the present study, many participants described the different ways they had

responded to and coped with the challenges attributed to CL/P. For those identifying as having adjusted positively to CL/P, there was a strong sense of determination to 'prove other people wrong' and to not let CL/P 'hold them back' in life. Such an attitude has been described as advantageous by other studies in this area (Meyerson, 2001; Strauss and Fenson, 2005; Egan et al., 2011). Participants also described the importance of taking a pragmatic approach to difficulties. For example, several participants described having deliberately put themselves in situations where they knew they would be challenged, such as becoming involved in activities which required regular public speaking, or meeting new people. For those with less confidence, avoiding situations which provoked anxiety was still a regular occurrence in adulthood. These participants were more likely to express the belief that CL/P had limited their achievements in life, and were more likely to interpret other people's perceptions of them as negative, and to respond in a similarly negative way. The benefits of 'active' or 'approach' coping (as opposed to avoidance) have been discussed a number of times in previous literature (Broder, 2001; Baker et al., 2009; Berger and Dalton, 2011). Knowledge derived from previous studies and the current study proposes that helping patients to identify the situations in which they feel challenged and providing them with tools to help them overcome anxiety may be a useful technique for intervention (also see Clarke et al., 2013).

Social comparisons

Another key factor which may mediate the impact of sociocultural pressures on appearance concerns is the direction and suitability of social comparisons. Theories of social comparisons claim that individuals regularly engage in making comparisons of our strengths and weaknesses to those of others, so that we can ascertain our relative standing in a given social environment (e.g. Festinger, 1954). While 'upward' comparisons with those we perceive as being 'better off' than ourselves are considered less adaptive, 'downward' comparisons can have a positive effect on mood and wellbeing (Festinger, 1954). Several studies, including the present investigation, have found downward comparisons to be a helpful coping strategy in adjustment to visible difference (Egan et al., 2011) and to CL/P specifically (Stock and Rumsey, in press 2013). These findings suggest the need for a better understanding of how social comparisons may affect adjustment, as well as supporting the idea that it is important to make social comparisons which are realistic, such as to friends and family members, rather than to

airbrushed media images and celebrities (All Party Parliamentary Group in Body Image and the Central YMCA, 2012).

Acceptance

For many participants in this study, CL/P formed an important part of their identity. Participants explained that they had learned to accept their condition, and expressed this by refusing to 'hide' their CL/P from others, and by talking about their condition openly. Previous research has also demonstrated the benefits of acceptance in relation to visible difference (Egan et al., 2011). Some of the techniques used in Cognitive Behavioural Therapy, including third wave approaches such as Acceptance and Commitment Therapy (ACT), are aimed at reframing a situation which is largely uncontrollable. Some of the principles of these approaches may therefore be beneficial in helping to facilitate acceptance in the field of visible difference (for an example see Pearson et al., 2010).

Faith and spirituality

Having faith, or spirituality, is thought to provide an additional source of support and promote a higher sense of belonging (Sodergren and Hyland, 2000). This was certainly true for some of the participants in the current study, who felt their faith had 'transformed' their personality and given them a strong sense of identity later in life. Research investigating the role of faith/spirituality in adjustment to visible difference is limited. However, a number of previous studies in the area have identified faith as a potentially important variable (Stock and Rumsey, in press 2014; Egan et al., 2011; Meyerson, 2001). An understanding of the ways in which spiritual beliefs and ideas may influence adjustment to a visible difference and possible ways of integrating some of this knowledge into intervention may thus be useful.

Dispositional style

Another factor which has been identified previously within the visible difference literature is that of dispositional style. As was apparent in the present study, adopting a more optimistic outlook towards life, looking at events in context and utilising a sense of humour in a positive way are associated with psychological adjustment and better overall health (Meyerson, 2001; Carr, 2004; Appearance Research Collaboration, 2009; Egan et al., 2011). Some researchers believe that optimism can be taught (Seligman, 1999), indicating that such

techniques could be integrated into psychological intervention for CL/P. Nonetheless, the findings of the present study imply that personality factors are complex, and further research is needed in order to understand the specific links between dispositional style and appearance-related adjustment.

Recognising positive growth

Within the present study, several participants believed that CL/P had shaped the person they had become and that this had largely been a positive experience. Many were able to identify the positive outcomes of growing up with CL/P and the opportunities they believed would not have occurred had they been born without CL/P. Participants also believed that having CL/P had helped them to manage other difficult events in their lives. Again, the importance of acknowledging positive growth in adjusting to conditions such as CL/P has been identified in previous literature (Meyerson, 2001; Strauss, 2001; Strauss and Fenson, 2005; Nelson et al., 2012b). The current study highlights the need for future research to include measures of positive growth, in order to aid our understanding of how this factor may contribute to adjustment.

Synthesis of findings

The aim of this study was to identify possible factors and processes contributing to psychological adjustment to CL/P from the patient perspective. 'Background' factors included age, gender, sexual orientation, the presence of additional conditions, socioeconomic status and adoption. 'External' factors included treatment autonomy, management of treatment expectations, familial appraisals, coping and support, salience of the cleft, public awareness of CL/P, degree of psychological input and opportunities for peer support. 'Internal' psychological factors included those related to appearance, such as perceptions of 'difference', perceptions of CL/P noticeability, internalisation of beauty ideals and appearance valence; factors related to social experiences, such as perceptions of teasing/bullying and social confidence; and factors related to the individual's outlook, such as a focus on strengths rather than deficits, responding to challenges, making appropriate social comparisons, acceptance, faith and spirituality, dispositional style and the ability to recognise personal growth.

Individual variation

Individual variation among participants was substantial, often resulting in positive, negative and neutral responses to the same concepts. This underlines 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 (e.g. age, gender, societal pressures, optimism, faith/spirituality etc.). These findings make it difficult 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 how these challenges may interact. Nonetheless, the findings of the present study resonate with the view that CL/P can be usefully conceptualised as an underlying stressor which calls upon an individual's energy reserves and coping resources to varying degrees throughout life (Lansdown et al., 1997).

Implications for research and clinical practice

This study identified several factors which may contribute to psychological adjustment according to the perspectives of individuals who have grown up with CL/P. 'Background' factors such as age, gender and ethnicity appear far from straightforward and warrant further investigation. Other background factors, such as sexuality, the presence of additional conditions, socioeconomic status and adoption have not been explored in relation to CL/P in great depth and represent a number of previously neglected and potentially vulnerable patient groups. Although background factors are largely contextual and thus unlikely to change, an increased awareness of the impact of such factors on adjustment to CL/P would allow for identification of those potentially at risk, improvement in the facilitation of appropriate support and intervention, and for appropriately targeted allocation of resources.

A number of 'external' factors were also identified. Although individual interpretation of the environment appears to play a key role, such factors are largely independent of the individual and may thus require a different level of intervention. Delivering basic training for health professionals, providing families and individuals with appropriate psychological support and information, and increasing general awareness of the conditions which affect appearance are not new ideas; however they are consistently raised within the CL/P literature. Efforts must be doubled in order to improve our understanding of these factors, to integrate this knowledge into practice, and to evaluate the impact of interventions in this area. National campaigns which tackle appearance-related stigma and promote diversity in appearance (see www.changingfaces.org.uk/Face-Equality for an example), school-based interventions which challenge sociocultural beauty ideals and increase media literacy (see Diedrichs and Halliwell, 2012), and a stepped approach from basic to specialist provision of

psychosocial support for appearance-related concerns within the health care system are also needed (see Clarke et al., 2013).

The 'internal' psychological factors reported in this study represent the many cognitive processes which may be involved in adjustment to CL/P and other visible conditions. Promisingly, these factors appear to be amenable to change. Furthermore, the challenges related to providing evidence of the worth of psychological interventions for this population are now well documented (Bessell and Moss, 2007; Normal et al., in press 2014; Jenkinson et al., under review). The factors identified in the current study, and how they may change over time, can potentially be measured. Thus, it is hoped that the findings of this study may offer some guidance for the development of new interventions, and for ways of assessing the value of interventions. A future challenge will be 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 is essential. The Cleft Collective, a UK-wide multidisciplinary prospective cohort study, represents one such opportunity to enhance our understanding of psychological adjustment to CL/P (Stock et al., under review).

The findings of this study resonate with the conceptual framework of adjustment to visible difference developed by the Appearance Research Collaboration (ARC; see Appearance Research Collaboration, 2009; Clarke et al., 2013). This framework has instigated a number of novel research projects, has proved useful as a guide for psychological interventions in this field and is applicable across a wide range of appearance-altering conditions (Clarke et al., 2013). The 'framework' approach provides the opportunity for consideration of a wider range of contributory factors than may be achieved through the majority of singular theories or models, and may thus offer a useful way forward for interventions in CL/P and in other fields. The present study has identified a number of additional factors which are not included in the ARC framework. This is likely to be due largely to the in-depth qualitative approach employed in the present study. Another speculation could be that some of the factors identified are specific to CL/P; however, existing knowledge of other appearance-altering conditions would anticipate similar findings across situations. The findings of the present study may provide additional insight and add to the ongoing development of the ARC framework.

Study limitations

Some limitations of this study must be acknowledged. First, the factors described in this paper have been identified through the subjective narratives of participants, who are attempting to find meaning in their experiences and which are open to the subjective nature of interpretation. Although inferences have been made about the characteristics of those who adjust well and those who experience difficulties, statistical evidence cannot be provided and causality cannot be assumed. Conversely, this approach can also be considered advantageous, since it provides a more in-depth, reflective and individual perspective which is rarely captured by quantitative methods alone.

Since all participants were self-selecting, this sample was open to bias. It is likely that those with a particularly negative or positive experience are more motivated to share their story. However, this largely did not seem to be the case with the current sample, since such a wide range of experiences were reported. While the sample did include a minority of participants with mental health difficulties, physical health problems, learning difficulties, dyslexia, hearing difficulties and/or lower SES, those who participated in the study may have been 'higher functioning' overall. Previous research with adult participants has commented on the need to include lower functioning groups in order to provide a full understanding of psychological status (e.g. Mani et al., 2010; Foo et al., 2012; Roberts and Mathias, 2012). Thus, the findings of this study must be interpreted in light of this possibility, and future research should attempt to recruit from appropriately diverse settings.

Through widespread advertising across the UK, efforts were made to include a large number of people from all backgrounds, of all ages and living in all regions of the UK. Nonetheless, it is apparent that some 'hard to reach' groups, such as those from different cultures and ethnic populations may not have been adequately represented. While it is common for those from different cultural communities to be under-represented in health research, particularly in the UK and specifically when researching a potentially sensitive topic, it is important to include such groups to address the health inequalities gap (Patel, 2013). Inclusion of diverse cultural groups is a frequently quoted area for future research and thus novel recruitment techniques, such as community networking and snowball sampling techniques (Patel, 2013), must be employed in future studies to increase participation rates.

Conclusions

This study has contributed knowledge of the possible risk and protective factors and processes contributing to psychological adjustment using a qualitative and patient-centred approach. 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 may offer guidance in relation to assessing the value of psychological interventions. There is 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 illustrate the potential degree of individual variation and offer insight into the conflicting results found within current literature. Finally, it is clear that much can be learned 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.

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