

“It doesn’t all just stop at 18”. Psychological adjustment and support needs of adults born with cleft lip and/or palate.

Abstract

Background: A cleft in the lip and/or the palate (CL/P) is considered to be a lifelong condition, yet relatively little is known about the long-term outcomes for patients. Existing literature is largely outdated and conflicted, with an almost exclusive focus on medical aspects and deficits.

Objective: To explore the psychological adjustment and possible support needs of a large number of adults born with CL/P from their own perspective.

Design: Fifty-two individual telephone interviews eliciting qualitative data.

Results: Qualitative analysis identified five themes. Participants reported a range of challenges in relation to ‘discharge’ from the service, additional surgery as an adult, social and romantic relationships, higher education, vocational achievement and access to psychological support. The findings imply that most adults with a cleft adjust well to these challenges and report many positive outcomes. For a minority of patients, issues attributed to the cleft may continue to cause distress in adulthood.

Conclusions: Adults with CL/P may require psychological support, information about the heritability of cleft, signposting and referrals from non-specialists, support regarding further treatment and opportunities to take part in research and activities. New issues arising in adulthood, such as entering the workplace, forming long-term relationships and starting a family, may warrant both further investigation and additional support. Further work is needed to identify the factors which contribute to psychological distress and resilience, as well as the timing of particular points of risk and opportunity for personal growth.

Keywords: cleft; adult; psychosocial; treatment; education; employment; quality of life; resilience

Introduction

A cleft in the lip and/or the palate (CL/P) can present a number of challenges for those affected. Such challenges may include treatment burden, dissatisfaction with appearance, problems with speech, impaired hearing and difficulties integrating with peers (Hunt et al., 2005; Rumsey and Stock, 2013). Although research in this area has focused predominantly on support for children and parents, a smaller number of studies have investigated the longer-term impact of CL/P.

Satisfaction with treatment

In a study by Sinko and colleagues (2005), adult patients with CL/P rated their aesthetic outcome to be significantly worse than when judged by a panel of independent experts. Of the seventy patients involved in the study, 44.3% asked for additional treatment. In several further studies, adults with CL/P were found to report less satisfaction with the facial features affected by the cleft, including the upper lip, nose, teeth and facial profile (Noar, 1991; Oosterkamp et al., 2007; Chuo et al., 2008). Similarly, Heller et al. (1981) reported a high rate of persistent dissatisfaction with appearance, hearing, speech and teeth in adults born with CL/P. Again, a high proportion of patients with CL/P requested further aesthetic and functional treatment (Ramstad et al., 1995b; Marcusson et al., 2002; Chuo et al., 2008).

Mental health

It has been suggested that the desire for further treatment is associated with poorer mental wellbeing (Sinko et al., 2005). Ramstad and colleagues (1995b) found anxiety, depression and palpitations to be reported twice as often by adults with CL/P compared to controls. These problems were closely associated with dissatisfaction with appearance, speech and dentition, and a desire for further treatment. In a study by Marcusson and colleagues (2002), dissatisfaction with appearance was significantly associated with depression and a desire to have additional operations. However, the opposite was also true; satisfaction with appearance was correlated with increased wellbeing in both adults with CL/P and the control group. Self-esteem has also been found to be lower in adults with CL/P (Berk et al., 2001; Cheung et al., 2007). Christensen and Mortensen (2002) reported that the risk of hospitalisation for mental ill health may be higher in patients with cleft palate only.

Social integration

Marcusson et al. (2001) found a marked impact of the cleft on adults' social life, while Berk and colleagues (2001) found adults with cleft to have less social support and higher levels of social anxiety compared to siblings without CL/P. Cochrane and Slade (1999) identified difficulties with social interaction, a negative effect on relationship formation and increased levels of social distress in adults with a cleft. These experiences were linked to a number of behavioural consequences, including withdrawal, avoidance and aggressive behaviours. Adults with CL/P have also reported experiencing negative reactions and discrimination in social settings (Sarwer et al., 1999; Patel and Ross, 2003). In a survey of 15- and 20-year-olds, 73% of patients with CL/P believed their confidence had been significantly affected by their cleft (Turner et al., 1997). Young adults with cleft may have fewer friends than their peers without CL/P (Ramstad et al., 1995b) and may struggle to initiate romantic relationships (Noar, 1991). A number of studies have also suggested that adults with CL/P are less likely to marry, marry later in life and are more likely to be childless (McWilliams and Paradise, 1973; Peter and Chinsky, 1974; Bjornsson and Agustsdottir, 1987; Ramstad et al., 1995a; Danino et al., 2005; Yttri et al., 2011). For those adults who are less well adjusted to their own experiences of CL/P, starting a family may evoke further difficulties (O'Hanlon et al., 2012; Stock and Rumsey, in press).

Educational achievement and work performance

Although no differences between adults with and without CL/P have been found in relation to employment status, a number of studies have identified lower levels of income, lesser scholarly and professional attainment and poorer job stability in adults with CL/P compared to controls (Peter and Chinsky, 1975; Danino et al., 2005; Ramstad et al., 1995a). Retrospective studies of school graduates have suggested that young adults with cleft may receive lower average grades and be less likely to receive a school leaving certificate than the general population (Persson et al., 2012). Recent neuropsychological studies have also suggested that adults with cleft may have significantly lower IQ scores and poorer cognitive functioning compared with matched controls (Nopoulos et al., 2002). Intellectual deficits have been particularly linked to individuals with palatal involvement (Eide et al., 2006).

Quality of life

Studies investigating overall quality of life (QoL), health-related quality of life (HRQoL) and oral health-related quality of life (OHRQoL) in adults with cleft have suggested that a number of dimensions may be negatively affected. These include social functioning (Sarwer et al., 1999; Marcusson et al., 2001; Sinko et al., 2005), physical functioning (nasal breathing, eating and speaking; Oosterkamp et al., 2007; Munz et al., 2011) and emotional functioning (Sarwer et al., 1999; Marcusson et al., 2001; Sinko et al., 2005; Foo et al., 2012). Cochrane and Slade (1999) identified seventy-five percent of adults in their study to have current difficulties which they attributed to their cleft. Some studies have identified potential differences in adult adjustment to CL/P according to age, gender and ethnicity (Ramstad et al., 1995b; de Sousa, 2008; Mani et al., 2010; Hutchinson et al., 2011; Reekie, 2011; Hamlet and Harcourt, in press), although these results are largely inconsistent. In a recent meta-analysis, Correa and colleagues (2014) concluded that adults with CL/P had lower scores on HRQoL compared to those without CL/P, with domains of psychological health and vitality particularly affected.

Positive outcomes and resilience

In stark contrast to some of the findings described above, a number of studies have found adults with CL/P to report levels of psychological wellbeing which are equal to or higher than their peers without cleft. Chetpakdeecheit and colleagues (2009) found young adults with cleft to have higher self-esteem and a greater capacity to cope with their social lives than controls. Cochrane and Slade's study (1999) indicated an elevated satisfaction with life, high positive affect and a number of positive consequences of having a cleft in adults, including strength of character and empathy for others, while Christensen and Mortensen (2002) found no evidence that the psychosocial stressors associated with CL/P and its treatment have a substantial impact on risk for psychiatric disease in adulthood. In a number of studies, many adults with repaired CL/P reported a high level of satisfaction with the treatment they had received, and with their overall facial appearance (Clifford et al., 1972; Bjornsson and Agustdottir, 1987; Noar, 1991; Semb et al., 2005). In a study investigating twenty South African adults, Patel and Ross (2003) found adults with CL/P to be generally satisfied with their communicative abilities, educational attainments, employment status and marital relationships. Having a child (with or without a cleft) may also offer adults with opportunities for personal growth and a chance to reappraise their experiences in a more positive light (Cochrane and Slade, 1999; Stock and Rumsey, in press). A number

of studies have thus concluded that in adults with CL/P, overall adjustment is similar to that of the general population (Bjornsson and Agustsdottir, 1987; Marcusson et al., 2001; Mani et al., 2010; Pisula et al. 2014).

Methodological challenges

While some of the variation in these findings can undoubtedly be explained by the complexity of psychological adjustment, contradictory findings may also be due in part to methodological challenges. Findings often vary according to the approach used, the measures employed, the comparisons (or lack thereof) made to appropriate control groups and the focus of the study (Klassen et al., 2012; Rumsey and Stock, 2013). Available samples are frequently small and heterogeneous in nature, and often do not take additional conditions and syndromes into account (Feragen et al., in press).

Existing literature on adults with CL/P has focused almost exclusively on medical aspects of care rather than patient experience, on objective measures rather than subjective self-perceptions and on deficits rather than strengths. Research in this field has shown little agreement to exist between patients and professionals in relation to surgical outcomes (Vargo et al., 2003; Foo et al., 2011; Alansari et al., 2014). Equally, the lack of association between the objective severity/asymmetry of the cleft and the patient's psychological wellbeing has been repeatedly demonstrated (Moss, 2005; Ong et al., 2007; Brown et al., 2010; Feragen et al., 2010). Further, few studies have offered suggestions of how to support this group of patients in practice.

A great deal of the available literature on adults with CL/P is now outdated, including much of that cited above, and must therefore be interpreted with caution (Roberts and Mathias, 2012). Continuing improvements in surgery and psychosocial care have led to better outcomes (Tatum and Losquadro, 2008). In addition, societal attitudes towards visible difference and disability may have changed over time (Hebl and Kleck, 2000). While researchers in this field initially hoped to draw attention away from the 'form' and 'function' of CL/P and toward the importance of the potential psychological and social impact, more recent research has been directed toward support and intervention, and to a more balanced view of both difficulties and strengths. Hence, modern research has emphasised the importance of incorporating measures of positive growth and resilience into studies of psychological adjustment to visible difference (Eiserman, 2001; Strauss, 2001; Egan et al., 2011). A number of further studies have proposed that the use of qualitative approaches may provide additional insight

into this complex field of research (Cochrane and Slade, 1999; Nelson, 2009) and help to shift the focus of care to one that is driven by a patient-centred agenda (Alansari et al., 2014).

Summary

Despite the many thousands of adults living with CL/P, both in the UK and around the world, research focusing on the psychosocial adjustment of this group is significantly lacking. Much of the existing literature is outdated, marred by methodological limitations and largely focused on faults and flaws. Further, there is a particular paucity of research exploring patients' self-perceptions. Nonetheless, it is clear that some adults may experience ongoing difficulties in relation to CL/P which could warrant further multidisciplinary support and intervention. Qualitative approaches may offer a unique perspective on this important topic and may help to address the issues which matter to the patients themselves. It is hoped that the findings of this study will have direct relevance for those adults who have already been discharged from the cleft service, as well as those children who are currently engaged in the revised treatment pathway.

Research aim

The aim of this study was to explore the psychological adjustment and possible support needs of a large number of adults born with CL/P from an in-depth qualitative 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 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. 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; $n = 6$). 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. The majority of 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 participants, 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 university press release.

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 encouraged to relay their own story. Discussion topics were chosen on the basis of previous literature and knowledge of this field. 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.

Ethical considerations

Ethical approval was obtained from the University of the West of England Research Ethics Committee. The study was also reviewed by the Cleft and Craniofacial Clinical Studies Group in the UK, which consists of clinicians, researchers and patient representatives. The research adhered to the British Psychological Society's Code of Ethics and Conduct (BPS, 2009) at all times. 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 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

Thematic analysis identified five themes: service transition and subsequent access; views and experiences of additional surgery; social and romantic relationships; higher education and vocational experiences; and psychological health and support. Each theme is described below with exemplar quotes. Pseudonyms have been used to protect participants’ anonymity.

THEME 1: Service transition and subsequent access

Participants in this study had been aged between 15 and 21 years when they were discharged from the cleft service. For many participants, this transition had been difficult.

There is a bit of a feeling of ‘oh, you’re 18 now, so thanks and good luck!’...but the treatment is ongoing really... When you’re 15 and you’re leaving the hospital you don’t think ‘wow, I’ll be 46 and this will still be going on’ – Julian, 46, UCLP

Once you get discharged it’s really difficult to go back to the doctor and say ‘I know I’m supposed to be an adult now, but I still need looking after, I still need to sit and ask questions’ – Fiona, 29, UCLP

For others, ‘leaving’ the cleft service had been a positive step.

Treatment was no longer in the equation and I felt that a good part of it was behind me...it was a form of closure really – Lucy, 36, BCL

Nonetheless, all participants in this study felt that the information available to adults, and ways of accessing this information could be improved. Many participants were unaware of their entitlement to treatment as an adult, and did not know how to regain contact with the relevant HPs. Some had paid for private treatment.

I didn’t know there was anything out there, there is no follow-up... I wouldn’t have a clue where to start... It would be nice to get updates or something from the cleft team, you know, you’re a member for life – Heather, 49, UCLP

I may just want a bit of emotional support...or there may be some new surgical technique or procedure...but I wouldn’t know because I’m out of the loop – Abigail, 33, BCLP

My local dentist just wasn’t specialist enough... In the end I had the bridge in my mouth done privately, and at great cost... – Henry, 59, UCLP

Participants had been disappointed not to be offered basic information and referrals by non-specialist HPs.

There are times when I’d like to know practical things, like when I go to my local GP or dentist...for them to say ‘actually I’m aware of this procedure’ or ‘have you thought about getting this done?’ – Julian, 46, UCLP

Participants also discussed the need for information and support regarding the heritability of CL/P.

I wouldn’t be worried about having a child with a cleft because I don’t think it’s a big deal...but there is a lot of work involved and it would be nice to be prepared – Natasha, 26, UCLP

These results suggest that ‘transition’ from cleft services and subsequent access to treatment may need to be assessed. Information regarding further treatment options in adulthood and the heritability of CL/P would also be valued.

THEME 2: Views and experiences of additional surgery

A number of participants had undergone further functional ($n = 7$) or aesthetic surgery ($n = 12$) as an adult. This had included orthognathic surgery and orthodontics, palatal closure or modification, lip revision and rhinoplasty. For some of these participants, these additional procedures had a positive impact.

The cleft team were fantastic...I was home in a couple of days and healing well and they were pleased with the progress...it was definitely worth it, despite the pain and discomfort, you have to keep thinking long-term –

Leah, 35 BCL

For other participants, these procedures had not been as worthwhile, or had been experienced as very negative.

There's no way I'd have any more surgery now...the grief that was involved surpassed the outcome really...it's a lot to put yourself through and in the end they are just tinkering – Kate, 35, UCLP

My experience of surgery is that it doesn't always work...it was quite traumatic really – Spencer, 46, UCLP

For two participants, undergoing surgery had provoked an unexpected emotional reaction.

I had some revisional surgery last year and the surgery itself was fine, but from then on everything went a little bit wrong and I ended up needing support from the clinical psychology team...[having surgery] brought up a lot of childhood issues...far more than I had anticipated... If I'd had more psychology input earlier on then I probably wouldn't have even considered [surgery] – Chloe, 37, BCLP

A number of participants reported feeling guilty or perplexed over having further surgery.

You can feel very, as if maybe you're doing it for the wrong reasons, you're doing it more for cosmetic reasons as an adult... 'is this the right thing to do or is this me being a bit picky?' – Lucy, 36, BCL

Others discussed the benefits of this opportunity.

People say to me 'you don't need anything done, I don't notice it', but it's me who's got to live with it and look in the mirror... Surgery now is a lot more successful... If it makes me feel better and they're willing to do it...then I should take it – Elizabeth, 52, BCLP

Many participants who had not yet undergone further treatment did not feel that additional surgery was necessary.

I think because I had a good repair in the first place...my scar isn't really that noticeable...I don't think they could do much to make it better to be honest – Mark, 27, UCLP

I thought about having more done, but I've lived with it for this long... I'm happy with how I am now really – Lauren, 58, UCLP

These findings suggest that additional surgery as an adult can have both a positive and negative impact. Clear and realistic information is needed, in addition to good communication between patients and health professionals, in order to manage patients' expectations.

THEME 3: Social and romantic relationships

For the majority of participants, friendships did not seem to be negatively affected by having a cleft.

My friends have always just accepted me for who I am, it's never been an issue... I don't think half of them even know I've got it – Kate, 35, UCLP

Others felt they responded differently to social encounters depending on the situation.

If I'm honest about it I think I can change between being quite shy and withdrawn to being pretty loud, as a way of making up for it and distracting people...depending on how I feel at the time and who I'm with – Natasha, 26, UCLP

Participants reflected on the lack of peer support they had while growing up. Several participants believed that meeting another adult with a cleft had been extremely beneficial and would be keen to offer support to others affected by cleft.

The first time I met someone else with a cleft was two weeks ago - and I'm 49! It was amazing just sitting there and finding out she'd been through the same things as I had – Heather, 49, UCLP

I would like to talk to parents that have just had a baby...or see other children with cleft and say 'it does get better' ... I think I'd be a good representation of the fact you can do anything...it doesn't have to hold you back
 – Teresa, 31, UCLP

Others commented that they would prefer not to engage in peer support.

Because it doesn't affect me in any way at all...I don't feel the need to bond with others with the same problem
 – Madelyn, 44, CP

In this study, 30 participants were in a long-term relationship at the time of interview. A further 17 participants identified as single, while four participants were either separated or divorced. One participant was a widower.

For most participants, CL/P had not had a great impact on romantic relationships in adulthood. Some participants commented that their cleft had affected their confidence in their early adult years.

In my late teens and early twenties I was very self-conscious about being rejected and I thought I couldn't have relationships in the same way as my friends, because I might not be attractive to the opposite sex... I did get very low at that time, because I thought 'well no one's gonna want me' – Robert, 60, BCLP

Others believed that having a cleft had made them less concerned with their appearance and more aware of what they wanted from a relationship.

I was quite a late starter, but out of choice...if a guy was rude to me in a bar I would just think 'well, you're an idiot then!' and walk away...I think a lot of my friends got in and out of bad relationships but I was happy being on my own and a bit more savvy about what I wanted from a relationship... You realise that people like you for who you are, not what you look like, and I think maybe you realise that earlier on – Carrie, 41, UCLP

Twenty-four of the adults who took part in this study were also parents. The remaining participants who had not yet started a family reflected upon the possibility of having a child with a cleft. Only two participants stated that this prospect would prevent them from having children.

It's not easy by any stretch of the imagination to have a child with a cleft because of the amount of 'extra work' ...but the only reason I would get rid of a child would be because there was something which would make

their life unbearable...that their quality of life was poor. And for me, cleft lip, cleft palate, that's not it – Fiona, 29, UCLP

On the whole, social and romantic relationships did not appear to be affected in this sample of adults. Young people may profit from psychological input focusing on social skills and intimacy, and some adults may benefit from receiving and providing peer support.

THEME 4: Higher education and vocational experiences

Forty-seven of the 52 participants were in employment at the time of interview. A further two participants had retired, and one participant identified as disabled. Two participants were unemployed due to mental ill health.

When reflecting on the impact of CL/P on their experiences of higher education and employment, many participants believed that having a cleft had not made a large difference.

I did my GCSEs, A Levels, did my degree and now I'm doing my Masters, and I'm a lead nurse, so having a cleft...has made no difference in terms of my education, I'm quite sure about that – Chloe, 37, BCLP

I've never been out of work. I'm in a managerial position now, so it hasn't really impacted on my life financially or intellectually – Liam, 55, BCLP

However, a number of participants described being faced by challenges in their educational and working lives. Some participants had found the tutors working in higher education to have a lack of understanding of CL/P.

Even when I was in university, there was a real thing where the tutors would look at you and think you were mentally slow...I think they expected less of you – Elijah, 32, UCLP

Others had been concerned about facing stigma when looking for employment.

Trying to get a job, that was always a big issue... I had a good degree and I went off to find work and in effect I was told my face didn't fit – Samuel, 44, BCLP

In my early twenties I set up a business with a university colleague...and the main investor said to my colleague that he wasn't sure he should invest in a company with one of the directors 'looking like he does' – Jason, 37, BCLP

The bit that I'm nervous about is not...whether I fit the criteria for the job, it's whether they're going to be able to understand me...and if they're going to accept me because of my scars – Fiona, 29, UCLP

Several participants discussed feeling as though they had to work much harder than their peers to prove they were able to achieve at a similar level.

I always felt that I had to be a little bit better than other people in order to compete for the job in the first place...and to work that much harder to prove that you are capable of doing the job once you've got it – Maria, 70, UCLP

Some participants believed that having a cleft had influenced their educational and vocational experiences in a positive way, and vice versa.

I started working in call centres where it was all about using my speech, which was my biggest hang up. I learned the phonetic alphabet, I learned how to be more confident over the phone...there was a definite positive shift in how I felt as an individual – Elijah, 32, UCLP

[My cleft] actually makes me more memorable...I think sometimes people warm to me because I've got it, because I'm someone who's slightly different being successful in what they do – Henry, 59, UCLP

The majority of participants in this study did not feel that having a cleft had held them back in relation to higher education and vocational experiences. However, these findings imply that a lack of awareness of CL/P in the general population may be a key factor for adults entering university and the work place.

THEME 5: Psychological health and support

Of the participants in this study, 30 identified as having adjusted positively to CL/P.

As a man of nearly 55 it doesn't bother me at all, I'm not self-conscious about the way I look or speak...I've got great friends, three lovely children...I've been married for fifteen years... I don't think it's done me a great deal of harm to be honest – Liam, 54, BCLP

A further thirteen believed that CL/P had an ongoing negative impact on their lives, with eight of these participants having disclosed previous or current mental health difficulties. This included clinical depression, anxiety disorders, Body Dysmorphic Disorder, eating disorders and difficulties with self-esteem.

It sounds dramatic but I feel like my whole life has been ruined by this...I've had lots of opportunities...things that I've desperately wanted to do, and I haven't done them, because of this feeling of being flawed...of not being good enough...and it all comes from the cleft – Louise, 45, UCLP

I thought I couldn't make myself prettier, so I'll make myself thinner, and I had anorexic tendencies... I thought 'one day I'll be prettier and it'll all be fine'...I still see it as a barrier – Grace, 28, UCLP

The remaining participants commented that difficulties relating to CL/P became more or less salient over time and in different situations.

It's a lifelong thing...you go through spells where you don't even think about it, and then something changes...and it maybe becomes a bit more of an issue for a while – Lucy, 36, BCL

Many participants did not feel they needed any psychological support as adults.

It's just something that I've never really talked about, not because I don't want to but because I don't feel there's a need – Hollie, 59, CPO

Others believed that receiving professional psychological support had been very beneficial.

Seeing the psychologist as part of the cleft team has made a massive, massive impact and it's completely turned round the way I see myself and where I go from here – Chloe, 37, BCLP

Participants identified a range of positive outcomes as a result of having a cleft. Many believed that the experience as a whole had been a positive one.

In terms of my development as a person, my character, the whole experience has been positive rather than negative – Anthony, 48, UCLP

These findings illustrate the complexity of psychological adjustment to CL/P, and highlight that access to psychological support is still valuable in adulthood.

Discussion

The aim of this study was to explore the psychological adjustment and possible support needs of adults born with CL/P. By conducting a large number of in-depth qualitative interviews with adults from across the UK, this study is able to offer insight into the psychological adjustment of this group of patients, as well as proposing a number of suggestions for future research and clinical practice. Implications arising from this study are discussed below, in relation to each theme.

Service transition and subsequent access

The participants in this study strongly articulated the view that access to information, treatment and support following discharge from the cleft service was of vital importance to adults. This need, particularly in relation to information regarding the heritability of CL/P, was raised in another recent study (Stock and Rumsey, in press). Following the reorganisation of cleft services in the UK, many cleft teams now take a lifespan approach to care. As a consequence, patients under the care of these teams are no longer ‘discharged’. Instead, patients are offered a multidisciplinary clinic appointment around the age of twenty years and encouraged to revisit the team (normally via General Practitioner referral) at any point in the future should they wish to. Referrals for genetic information, advice or counselling for those keen to know more about the heritability of CL/P can be made, but this is currently not part of the standard protocol for most teams. UK cleft teams may wish to consider the opportunity of raising this topic with young adults during patients’ 15- and/or 20-year appointments at multidisciplinary clinics. Additionally, it is important for non-cleft specialists (such as GPs and dentists) to be able to make appropriate referrals when necessary, since they are ideally placed to act as a conduit between the patient and the specialist cleft team. It is recommended that a lifespan approach to cleft

care be adopted by all healthcare providers where possible, to ensure that patients' ongoing information and support needs are met. For those patients who feel able to initiate contact when needed, a move away from scheduled appointments toward 'open access' services may be helpful.

Views and experiences of additional surgery

A proportion of the participants in this study had undergone additional aesthetic or functional surgery as an adult ($n = 19$, 36.5%). This finding is relatively in line with the percentage of adult patients who reported a desire for further treatment in previous literature (Ramstad et al., 1995b, 35%; Marcusson et al., 2002, 47%; Sinko et al., 2005, 44.3%). Among this literature, and in the present study, the desire for additional treatment seemed to be linked to dissatisfaction with appearance, as well as the belief that surgical technique had improved since participants were originally treated. Although some participants who had undergone additional surgery were satisfied with the results and found this process to be beneficial, other participants did not believe the benefits outweighed the risks. Some participants worried whether surgery was still 'necessary' and whether considering aesthetic revision was a question of 'vanity'. Other people's opinions of cosmetic surgery seemed to influence participants' treatment decision-making to some degree and exacerbate these feelings of confusion and/or distress. For two participants, re-entering the treatment system had evoked a strong emotional reaction which subsequently required psychological intervention. Some of the cleft literature has suggested that poorer mental wellbeing may be associated with a desire for additional surgery (Marcusson et al., 2002; Sinko et al., 2005). In the general cosmetic surgery literature, much attention has been paid to the 'reasons' behind wanting surgical intervention. Predictors of desire for cosmetic surgery in the general population include lower self-ratings of physical attractiveness, higher appearance investment, acceptability of cosmetic surgery among family and friends, past experience of surgery and susceptibility to media messages concerning appearance (Brown et al., 2007; Swami et al., 2008; Swami, 2009; Slevic and Tiggemann, 2010; Ashikali et al., in press). This literature may be useful when interpreting the results of CL/P studies. The findings of the present study point strongly to the importance of incorporating psychological support into treatment-based decisions and carefully managing adults' expectations of surgery. Integration of evidence-based psychosocial intervention which combats appearance dissatisfaction among individuals with cleft is key (e.g. Clarke et al., 2013).

Of those participants who stated they would not have any further corrective treatment, some were ‘fed up’ of having treatment (Sinko et al., 2005), while others were reasonably satisfied with their surgical outcome and accepting of their current appearance. Several participants believed their cleft was no longer noticeable to others, while many felt that appearance became less important as an adult. Research in the field of visible difference has shown that self-perceptions of noticeability account for far more of the variability in psychological wellbeing than objective ratings of severity/asymmetry (Moss, 2005; Ong et al., 2007; Brown et al., 2010; Feragen et al., 2010). In addition, the emphasis that a person places on their appearance relative to other personal qualities or characteristics is known to be important in relation to appearance dissatisfaction and psychological wellbeing (Appearance Research Collaboration, 2009). These findings illustrate some of the important factors involved in the development of appearance concerns and provide strong support for the inclusion of patient-rated measures of treatment outcomes.

Social and romantic relationships

Although previous studies have reported a marked impact on social life for adults with CL/P (McWilliams and Paradise, 1973; Peter and Chinsky, 1974; Noar, 1991; Ramstad et al., 1995b; Turner et al., 1997; Berk et al., 2001; Danino et al., 2005), the findings of the present study found the majority of participants to be socially well-adjusted and involved in long-term romantic relationships. However, some of the interviews alluded to a raised level of social anxiety in certain situations. Some participants had developed strategies to cope with this, such as withdrawal, or being overly ‘loud’ or ‘pushy’ to compensate for their discomfort. These types of behavioural responses in adults with cleft (and other visible differences) have been identified in previous literature (Macgregor, 1974; Cochrane and Slade, 1999, Appearance Research Collaboration, 2009). Some participants had also struggled to form romantic relationships during their early adult years; this was predominantly attributed to a lack of self-confidence. This supports findings by Turner et al. (1997) that patients’ confidence can be affected by the cleft and suggests that implementing psychosocial support for young adults in relation to social skills and intimacy may be beneficial (Bull and Rumsey, 1988; Kapp-Simon, 1995; Appearance Research Collaboration, 2009; Griffiths et al., 2012).

While several participants reflected that their confidence had delayed them from entering into a romantic relationship, some felt this had been a conscious choice. Others commented that romantic relationships had often developed out of existing friendships. It may be that being born with a condition which is visible to others can sometimes result in one placing less value on appearance, and more value on meaningful companionship.

Twenty-four of the 52 participants in this study had children of their own. A small minority of participants had consciously chosen not to have children, although only two stated that this was due to the recurrence risk of CL/P. Many participants wanted to have children in the future and strongly believed that being born with a cleft would not significantly affect their child's quality of life. Although previous literature has reported that adults with CL/P are more likely to be childless (Yttri et al., 2011), on the whole this did not seem to apply to participants in the present sample. It is likely that an individual's appraisal of their own experiences affects how they perceive the idea of becoming a parent, and the possibility of their own child(ren) being affected by CL/P (O'Hanlon et al., 2012; Stock and Rumsey, in press).

Although not all participants wanted to engage in peer support, many others had found meeting someone else with a CL/P to be extremely valuable. Engaging in mutual support can facilitate personal growth through shared experiences and be of benefit to both parties (Cohen and McKay, 1984). The number of adults in this study who had never met anyone else with a cleft was surprising. Offering patients with CL/P opportunities to meet others in a similar situation may be helpful for some, particularly if others are slightly ahead along the treatment pathway (Stock and Rumsey, in press). Many participants also felt they would be ideal advocates for others affected by CL/P, including parents and young people. They believed they could show others that having a cleft does not need to be restrictive and that the long term outcomes for people with CL/P can be very good. In addition, participants felt they could contribute to the understanding of CL/P through participation in research and related activities. Offering adults with CL/P opportunities to 'give something back' could help to reassure parents of the positive outcomes for their offspring with CL/P and provide young people with cleft with a unique and inspirational perspective.

Higher education and vocational experiences

The vast majority of literature investigating the educational and vocational attainment of individuals with CL/P suggests that those born with a cleft are likely to be educationally disadvantaged (Peter and Chinsky, 1975; Ramstad et al., 1995a; Danino et al., 2005; Persson et al., 2012). Again, this did not seem to be the case in the present study. Only three participants had been out of work (for reasons of disability). The remaining participants had achieved a high educational level (*n* of participants with higher education qualifications = 39) and were employed in a range of settings. A sampling bias may account for some of the discrepancy in these findings. It could be that those with a higher intellect were more likely to be aware of and/or chose to take part in this study. Another possibility is that only a small proportion of participants reported having an additional condition which may affect achievement, such as dyslexia or a learning difficulty (*n* = 5). A high prevalence of such conditions has been found among children with CL/P (Feragen and Stock, 2014) and is not often accounted for in quantitative research (Feragen et al., in press). Another possible explanation for this discrepancy could be that the ‘objective’ deficits described in the literature are not necessarily experienced subjectively by patients. In any case, it is of vital importance that clinicians are able to provide parents of children with CL/P with a realistic view of their child’s likely developmental trajectory.

Several participants had perceived some form of stigma to exist in higher education and employment settings. This had made some participants feel as though they had to work harder than their peers to ‘prove’ themselves capable. While some individuals may rise to this challenge and become more determined to succeed, others may feel discriminated against. On the whole, participants did not feel that having a cleft had negatively impacted on their level of academic or vocational achievement. Some believed their cleft had been a positive asset in the workplace, making them more ‘memorable’. Early studies of attitudes towards visible difference demonstrated that individuals who possess a high level of social skill elicit significantly more favourable impressions regardless of the presence or absence of a visible difference (Rumsey et al., 1986). Again, investing in the development of social skills in order to tackle some of the existing societal stigma may be of help to those affected by a visible difference (Bull and Rumsey, 1988; Kapp-Simon, 1995; Clarke et al., 2013). Further investigation of the educational and workplace environment in relation to CL/P is needed in order to understand more about the factors which contribute to academic and vocational achievement in individuals with a cleft.

Psychological adjustment and support

While most participants in this study believed they had adjusted well to having a cleft, some felt that cleft-related issues became more or less salient over time and across different situations. Others believed that CL/P had an ongoing negative impact on their lives, and attributed a number of current difficulties to their cleft. It is important to remember the lack of psychological support that would have been available to these participants while growing up. However, these participants represented a much smaller proportion than has previously been reported. For example, Cochrane and Slade identified seventy-five percent of their sample to have problems in adulthood attributed to CL/P. The notion that an individual will fall somewhere on a ‘continuum’ of adjustment explains some of the discrepancy in the existing literature (Appearance Research Collaboration, 2009). In addition, levels of adjustment have been shown to fluctuate in adults with visible differences (Appearance Research Collaboration, 2009). If we are to move forward, we need to be able to identify points of risk and opportunity for growth, as well as understand more about the factors which contribute to the development of psychological distress and resilience in individuals with cleft (Klassen et al., 2012; Rumsey and Stock, 2013). Participants in this study were able to identify a number of positive consequences of being born with a cleft, providing further support for including measures of personal growth and resilience in future research. The findings of the present study also illustrate the importance of early and ongoing psychological input for those affected by CL/P.

Methodological observations and study limitations

The findings of the present study seem to be more in line with the existing (albeit limited) qualitative research in this area, rather than the results derived from previous quantitative research. While this may be illustrative of the difficulties of capturing psychological adjustment using quantitative approaches, it must also be recognised that individuals who take part in qualitative research are offering a narrative which is constructed through their attempts to make sense of their view of the world. A number of adults who participated in the present study commented that if they were to be interviewed again in the future, they may have responded differently to some of the questions. Ultimately, longitudinal research is needed in order to understand the complexities and fluctuation of psychological adjustment to CL/P.

Very few of the adults who took part in this study identified themselves as being from an ethnic minority group. While this is not unusual in psychological research, efforts must be doubled if we are to provide an evidence base which is not limited by social and cultural factors. This study was unable to gauge any effect of age or gender; individual variation appeared to be considerable. Future quantitative research which builds upon qualitative findings may be able to better address these potential variables.

All of the adults who participated in this study were treated prior to the reorganisation of cleft care in the UK. Thus, the findings from this study have direct relevance for those adults who have already been discharged from the cleft service. As has been suggested by this study and other recent research in the area (Stock and Rumsey, in press), the best ways of accessing this sub-group of the CL/P population requires consideration. The findings of this study may also be used to inform current practice, with a view to adopting a preventative approach toward difficulties in adulthood for those children who are currently engaged in the treatment pathway. Finally, although the composition and configuration of cleft services varies significantly from country to country, many insights from this study are broadly applicable to different care contexts.

Conclusions

CL/P is a lifelong condition. For some, it represents an underlying stressor that becomes more or less salient over time and according to different situations (Lansdown et al., 1997). The findings of this research imply that most adults with a cleft adjust well to these challenges and report many positive outcomes. However, for a minority of patients, issues attributed to the cleft may continue to cause distress in adulthood. Adults with CL/P may require psychological support, information about the heritability of cleft, signposting and referrals from non-specialists, support regarding further treatment and opportunities to take part in research and activities. New issues arising in adulthood, such as entering the workplace, forming long-term relationships and starting a family, may warrant both further investigation and additional support. Further work is needed to identify the factors which contribute to psychological distress and resilience, as well as the timing of particular points of risk and opportunity for personal growth.

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