

## **Starting a family: the experience of parents with CL/P**

### Abstract

**Background:** One of the key challenges facing young adults with CL/P is decision-making around starting a family, due to the increased likelihood of their own child being diagnosed with CL/P. Should this occur, a second key challenge is how to deal with their child's diagnosis and subsequent treatment.

**Objective:** To explore the views, experiences and possible support needs of this unique group of parents in order to inform the services provided by non-specialist Health Professionals, cleft teams and genetic counsellors.

**Design:** Individual telephone interviews eliciting qualitative data.

**Results:** Qualitative Thematic Analysis identified five themes. Accessing accurate information and appropriate support around heritability presented a significant challenge. Parents described feelings of responsibility and distress at their child's diagnosis, as well as a number of factors which had helped or hindered their adjustment. Parents also described ways in which their own experiences had impacted on their parenting style, and how becoming a parent had changed the way they felt about their own cleft.

**Conclusions:** Young adults' understanding of what it means to grow up with CL/P may impact on their decision to start a family and their experiences of having children. Possible methods of supporting prospective parents through this potentially difficult stage will be discussed.

**Keywords:** cleft, adult, parent, genetics, psychological adjustment, qualitative

## Introduction

In the UK and around the world, thousands of adults are living with cleft lip/palate (CL/P). For the majority of patients with cleft, routine treatment is likely to conclude when they reach the age of eighteen years. Although CL/P is considered to be a lifelong condition, virtually no research to date has documented what happens to young adults once they have left the service.

One of the key challenges facing young adults with CL/P is decision-making around starting their own family (Patel and Ross, 2003; Williams et al., 2012; Roberts and Mathias, 2012). Although the genetic basis of clefting is not yet fully understood, it is known that the chance of having a child with a cleft is higher if one or both of the parents has a cleft themselves. If an individual's cleft has been associated with a known syndrome, the recurrence risk will be higher still, depending on the genetic variation of the syndrome identified (NHS National Genetics Education and Development Centre, online resource). Research with other heritable conditions has suggested that adults may choose not to have children on the basis of genetic risk (e.g. James, Hadley, Holtzman, Winklestein, 2006; de Die-Smulders, de Wert, Liebaers, Tibben and Evers-Kiebooms, 2013). However, little research has been conducted with prospective parents with CL/P.

Of the research that has been conducted with this unique group, one Danish study suggested that women who are born with CL/P are more likely to be childless, or to wait longer to have their first child when compared to the general Danish population (Yttri et al., 2011). However, not all research in the field of CL/P has echoed this finding (Andrews-Casal et al., 1998), with some prospective parents reporting positive views of having children regardless of recurrence risk (Patel and Ross, 2003). Nonetheless, the birth of a child can be an emotional and stressful time for any new parent, and the increased possibility of having a child with a health condition may impact to some degree on the process of deciding whether to have children.

A second key challenge for young adults with CL/P is how to deal with a diagnosis of cleft in their child should this occur. A wealth of previous research has demonstrated that for parents who do not have CL/P, receiving a diagnosis of cleft in their child can be very distressing (e.g. Nelson et al., 2011; Williams et al., 2011). Initially, parents are likely to feel shocked and daunted by the potential challenges that lie ahead, and throughout their child's development the burden of care can be considerable. Longer-term implications may include elevated

levels of parental stress, anxiety and depression, a risk of maternal detachment, lower cognitive functioning in the child and family discord (Drotar, 1997; Cohen, 1999; Collett and Speltz, 2006). The risk of such outcomes may be exacerbated if appropriate support is not readily accessible throughout the family's journey (Baker et al., 2009; Knapke et al., 2010; Vanz and Ribeiro, 2011; Nelson et al., 2012).

Within previous literature on this topic (albeit very limited), it has been suggested that parents who have a cleft themselves may experience more distress at the prospect of having a child with CL/P than those parents who do not have a cleft (Andrews-Casal et al., 1998; O'Hanlon et al., 2012). This is largely based on the assumption that having a child with a cleft will conjure strong feelings of guilt, and evoke parents' difficult memories of growing up with the same condition, impacting on their ability to cope. However, studies using quantitative methodology to explore this have consistently failed to identify significant differences in distress between groups of parents with and without CL/P (notably Andrews-Casal et al., 1998; O'Hanlon et al., 2012).

While reported levels of distress may not vary significantly between parents with and without a cleft, this type of approach does not provide any detail about the nature of distress experienced by parents with CL/P in relation to starting a family, nor does it address possible differences in the type of support that may subsequently be required. O'Hanlon et al.'s study included a qualitative component, which provided further insight. This aspect of the research suggested that parents with a diagnosis of CL/P may experience and cope with their child's diagnosis in different ways to those parents who do not have a cleft. This may explain some of the discrepancies reported in the data thus far and points to the value of exploring parents' experiences from a more in-depth and individual standpoint.

Within the wider genomics and healthcare literature, studies have demonstrated the need for appropriate intervention, psychosocial support and guidance for those with a variety of heritable health conditions (Miller et al., 2006; Harper, 2010). In the field of craniofacial research, few studies have discussed potential methods of support and/or intervention in relation to this key challenge faced by many young adults. An awareness of the views, experiences and possible support needs of this unique group of parents would thus offer a number of useful implications for clinical practice and future research. The present study aims to contribute to the small but important body of research in this area, with a view to informing the services provided by non-specialist Health Professionals (HPs), cleft teams and genetic counsellors. To the authors' knowledge, this is the first

study to conduct in-depth qualitative interviews with this distinct group of parents. This is also the first study to include parents with CL/P whose children were not born with a cleft.

## Method

### *Design and Participants*

In view of the limited results available within 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).

Parents were recruited as part of a larger study involving adults with CL/P (in preparation). Since a wealth of research has been carried out relating to the experiences of parents who do not have a cleft themselves, the current study focused solely on parents with CL/P. 24 parents with a diagnosis of CL/P participated. Of these parents, 8 had children who were born with CL/P, while 16 had children without CL/P. No substantial differences in demographics between these two groups of parents were identified. 13 of the parents who took part were mothers, while 11 were fathers. Parents were aged between 28 and 70 years at the time of interview (average 43 years). 22 parents identified as White British, while one parent identified as Mixed Caucasian and one as British Asian. The majority of parents were in a long-term relationship ( $n = 19$ ), and were employed at the time of interview ( $n = 22$ ). Parents had a range of different cleft types themselves, including UCLP ( $n = 12$ ), BCLP ( $n = 8$ ), CPO ( $n = 3$ ) and CLO ( $n = 1$ ). Twelve parents were recruited through the UK-based charity, the Cleft Lip and Palate Association (CLAPA), while the remaining twelve were recruited through other means (e.g. a university press release). Participants were recruited from across the UK, with representation from most regions. 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 parents in this study identified themselves or their children as having an associated syndrome.

Once informed consent had been obtained, individual, free-response recorded telephone interviews were conducted until saturation was achieved (twenty-four parents). 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 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. As this study was part of a larger research project exploring the experiences of adults with CL/P, topics which the interviewer aimed to cover included: family history of cleft; school experiences; treatment journey; transition into adulthood; decision-making around starting own family; experiences of having children; past and current support needs; and reflection of the journey as a whole. Interviews lasted between 40 minutes and one hour.

### *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. 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 both 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; these are outlined below using illustrative quotes. Although some of the points raised are more applicable to parents of children who were born with CL/P than to parents whose children were not born with a cleft, on the whole themes were found to resonate across both groups of parents. Data collected from all 24 parents has therefore been merged in the Results section below. Where minor differences were observed, these have been noted as appropriate. All participants have been given pseudonyms to preserve anonymity.

## 1. Issues surrounding heritability

At the time their child was conceived, almost half of all parents in this study had been unaware that CL/P can be a hereditary condition.

*Julian – I was in complete ignorance, I had no idea that there were any indications, the information wasn't there and nobody ever told me it could be a possibility.*

These parents commented that if they had been aware of the possible genetic link they would have had time to prepare.

*Julian – I think we might have thought, maybe we do need to take extra care during early pregnancy.*

*Aiden – It wouldn't have stopped me having children but it might have made me talk to somebody. Obviously I had a partner at the time so I would have said 'these are the risks' and passed that information on.*

Some parents had been given misinformation about the likelihood of their children having CL/P.

*Liam – I was told 'yours is just a one-off, it's something that happens occasionally and it's not hereditary'.*

*Hannah – When I found out I was pregnant the GP just said 'don't worry; it will probably skip a generation'.*

Even for those parents who had been aware of the potential heritability, the genetic mechanisms involved in clefting remained ambiguous at best.

*Jacob – It seemed to follow a pattern in my family...until my son was born, which blew all the statistics out the water.*

Some parents also commented that 'knowing the odds' of having a child with a cleft was not always helpful.

*Emma – For me, not knowing the exact odds is easier...they might say the risk is sixty per cent, but I'd still be worrying about the other forty per cent...it would just create a massive spiral in my head.*

Finding the relevant information, or knowing how to approach the topic with a Health Professional, had also been difficult for many participants.

Tom – *We never got the chance to talk to anyone about genetic screening and it was a bit unclear to us about how we could tap into that.*

Grace – *At my twenty week scan I said ‘is my baby’s face OK?’ and she kind of looked at me like I was stupid, and I was like ‘well I don’t know, no one’s ever explained it to me!’ I expected them to bring it up to be honest.*

For a small number of parents, the possibility of their grandchildren being affected by CL/P was also raised.

Samuel – *I wonder if maybe in twenty years’ time when my kids get married, will this be a problem for my grandchildren? That’s a bit of a weight on my mind.*

The degree to which heritability of cleft was reportedly misunderstood by parents with CL/P and Health Professionals is both significant and concerning.

## **2. Reactions to their child’s diagnosis**

Parents of children who had been born with a cleft reported a number of distressing reactions to their child’s diagnosis.

Sofia - *I was devastated. People say things like ‘oh there’s so much that can be done nowadays’...but I know how painful it is to get there, and how long the journey is. I just felt sad for her really.*

Lauren – *I thought, it’s going to be another twenty-plus years of cleft, of supporting him and helping him through it...I just wanted someone to say ‘I’m so sorry that you have to go through this again’...it was a very, very tough time.*

Equally, parents whose children were not born with CL/P had speculated on how they might have felt had their child received a diagnosis.

Henry – *I would have found it very difficult...originally I didn’t want children because of it. I would have always felt responsible in a funny sort of way, even though you can’t help it...but then again, there are so many things a child can suffer from, and I suppose cleft is a mild one really.*



Sean – *My cleft came out of the clear blue sky, so nobody could pinpoint it, but suddenly amidst all the scans it occurred to me that if this baby had a cleft it would be down to me. But when I thought about it more I realised 'is it such a terrible thing?' so I would say I was slightly conflicted at that point.*

However, not all parents reported feeling concerned about having a child with CL/P.

Ben – *It never played on my mind at all...When we found out it wasn't a disappointment or a shock, it was just 'OK, it is what it is and we'll deal with it'.*

Ethan – *It isn't an issue for me and never has been. I had such a good experience of life anyway that I kind of assumed I could do the same for my kids.*

The majority of those participants that had initial concerns subsequently felt they had been/would be able to adjust well to their child's condition.

Julian - *At the end of the day, I came out alright, and at a time when treatment wasn't as good, so it would be a hurdle but certainly not one we couldn't overcome.*

No parents in this study reported that they had considered terminating a pregnancy, and many had been upset by media reports of unborn children being aborted on the grounds of having CL/P.

Madelyn – *There was a case on the radio about a woman who wanted to abort a child because it had a cleft and I was really quite appalled.*

Parents whose children had not been born with CL/P often reported feeling 'lucky'. However, the possibility of having a child with a cleft in the future was still a concern for some.

Emma – *I was very, very lucky that she hasn't got a cleft, but if I did go down the road of looking to have more children I would obviously be back in that position again.*

It is clear from these findings that factors such as the parents' own experiences of growing up with CL/P may contribute to considerable differences in their response to their child's potential diagnosis.

### 3. Factors affecting parental adjustment

Parents of children with CL/P felt that having had children without CL/P previously, or receiving their child's diagnosis of cleft postnatally, increased the level of shock they had experienced following the birth of their child.

Lauren – *When we had our first son we did worry, but he was absolutely fine, so when we had our second son and he had a cleft...it was a bigger shock.*

Liam – *It didn't show up on any of the scans, so until the birth we thought we had gotten away with it, if you know what I mean.*

Some parents compared their child's cleft to their own, assuming that the level of treatment needed would be more or less depending on the type of cleft their child had.

Abigail – *She only had a unilateral, whereas I had bilateral, so that was a big relief to me, knowing hers wouldn't be as bad as mine.*

A lack of understanding of CL/P from family members, friends and members of the public seemed to impact significantly on the adjustment of some parents.

Lauren – *My father-in-law blamed me. My parents took it quite badly as well. My husband was very supportive but he hadn't been through it himself, so he didn't understand how long it would take and how much it involved. I don't think it registered in him in the same way as it did me, so it created a bit of a rift. There were a lot of emotions flying around at that time.*

Samuel – *Friends will say 'I can't believe your son's so handsome' and I don't quite know how to take that really.*

Abigail – *I feel quite self-conscious when I'm out with my daughter, I think we do tend to get more looks from other people when we're together.*

For a minority of parents, having faith, or spirituality, was reported to be a strong source of support, and shaped the way they felt about having children.

Lucy – *I think because of my Christian faith, the way that you're made and created, that's the way you're meant to be, so [my child] having a cleft would have been something I could have coped with.*

Parents commonly reported that non-specialist HPs had a lack of knowledge about cleft, and that comments from all HPs, including specialists, had been insensitive at times.

Liam – *I remember them delivering his head and the room went completely silent... Eventually my wife said 'what's the matter?' and I said 'he's got the same condition that I've got', and that was like popping a bubble, everybody went 'oh that's fantastic, so you know what to do'. It quite upset me at the time because I'd basically let them off the hook.*

Lauren – *At my son's first operation, the consultant actually looked up at me and said 'oh, well you know what it's like don't you?' and I was thinking 'yes, but I don't need to be reminded!'*

Following a diagnosis of cleft in their child, a small number of parents felt that being able to support other families of children with CL/P from a unique perspective had been helpful, although some felt they were relied on too much at times.

Liam – *After my son was born I was asked to come and speak to other parents. I would walk in and speak to them normally and demonstrate that I was normal in every way except I had a cleft. While it was a good thing, I think that support should have been supplied on a more formal basis, rather than just phoning somebody up who has the condition and saying 'can you talk to some [new parents]?'*

While parents with CL/P report similar occurrences to those reported previously by parents who do not have CL/P themselves, these occurrences appear to be more complex if the parent has their own experiences of CL/P.

#### **4. Impact of own experiences on parenting style**

For a small number of parents, watching their child go through operations brought back upsetting memories of their own treatment.

Lauren – *At the first operation I physically couldn't take him down to theatre...because I had so many memories of going under the anaesthetic myself. It was just too close to me.*

Several others felt that they were more relaxed about their child's treatment than parents who had no experience of cleft.

Ben – *I didn't really realise the impact of it until we had that session with the other prospective parents. It really shocked me and I felt a little bit annoyed that people were so worried about something that for me was just a very normal part of my life.*

Abigail – *There were parents in the hospital who were, this sounds harsh, but they were faffing around, whereas I was a bit more 'this is how it's going to be' ...and pragmatic. You know what's got to be done so you just do it.*

Almost all parents believed their own experiences would be of benefit to their child, whether or not their child had been born with a cleft.

Aiden – *I tell my daughter she's a bit like me, because she's had her lip fixed too...it's quite positive that I've gone through it, so she's got somebody that she can talk to and discuss any fears.*

Ben - *Because my father had a cleft as well, it's always been there, and it normalised it for me. I hope I can have that same positive effect on my son.*

Grace - *I think in one respect I would have been the best person to deal with it; I know the ins and outs of it and I wouldn't settle for less than perfect when it came to the care he would have...even though he doesn't have a cleft I make sure he understands not to judge people on their appearance.*

A minority of parents commented that having a child with a cleft had brought up some of their own unresolved issues, and that it had been difficult not to bestow these feelings onto their child.

Lauren – *I think even now there's probably still a lot there, that bubbles around. In a way I'm wanting to move forward, but it's also acknowledging what one's been through, and acknowledging what my son and I have been through together. It is so hard not to put your own emotions and fears onto them.*

Several parents commented that because of their own positive or negative experiences, they had made a conscious effort to avoid or repeat these experiences with their children. This was apparent irrespective of whether their child had CL/P or not.

Chloe – *We didn't talk much as a family when I was growing up; things were done differently in those days, so I make a real conscious effort to talk to my son, and be open with him.*

Sofia – *I hated moving schools when I was younger, so I'll make sure she stays in the same school. Also I think bullies thrive on people's insecurities, so I'll just try to make her as self-confident as she can be really.*

Other parents commented that they hadn't treated their children any differently.

Maria – *Just as my mother treated me, you treat them as if they're absolutely normal, that they can cope and that they can get on with it.*

Having a child (with or without a cleft) is likely to evoke parents' experiences of growing up and impact on the way they choose to parent their child. For some, the triggering of these memories may cause some distress.

## **5. Impact of becoming a parent on own outlook**

Several parents felt that having a child (with or without a cleft) had changed their feelings about having CL/P themselves.

Emma - *I think your focus shifts when you have a child. I have to think of her rather than myself, and it gives you a different perspective... I wouldn't have any surgery done now. My daughter knows me as I am, and I don't think I could do that to her; changing mummy's image and her feeling like 'well you're not the mummy I know'.*

Others had found that having a child had put them back in touch with the health service. In addition, seeing their child go through routine treatment had 'inspired' a minority of parents to have further treatment themselves.

*Olivia – My son actually inspired me; he doesn't have a cleft but his teeth didn't grow straight...he went from having really crooked teeth to this amazing Hollywood smile! He looked incredible and the change in his confidence was phenomenal; I think I wanted a bit of that. That's what really spurred me on to go and talk to my dentist and get myself back in the treatment loop again.*

A small number of parents commented that becoming a parent had given them a greater understanding of their own parents' experiences.

*Olivia – It's quite stressful as a parent watching your child go through anything, and as a mother now I can see how it would feel for my mother watching me.*

Several of the parents whose children had been born with CL/P commented on the uniqueness and strength of their relationship with their child, and the impact that this relationship had on their own ability to cope.

*Lauren – I think my son and I are very close, and I look back over the times we went to appointments and driving there in the car with him as very special times, it's very unique, and I wouldn't have had it any other way. My son has given me huge strength, just to cope. Through him I've learned a huge amount from the other side.*

Having a child (with or without a cleft) may alter parents' perspectives about growing up with CL/P. It may also prompt parents to seek further treatment, particularly if they perceive care to have improved.

## Discussion

This research provides unique insight into the views and experiences of adults with CL/P who go on to have their own family. A number of discussion points have been generated by the analysis of participants' responses

and are outlined below in relation to each theme. Findings and clinical recommendations are also summarised in Table 1.

### *Heritability of CL/P*

One of the strongest themes to be identified from this analysis was the lack of understanding in relation to the heritability of CL/P. Despite having been born with a cleft themselves, many parents had been unaware that CL/P could be passed on to their children. While public understanding of genetics may have changed during the last decade, this finding highlights the need to raise awareness of the issues surrounding heritability among patients with cleft. A number of parents were aware that having a cleft increases the risk of recurrence in their children, but had been unsure how to access the information they needed. Similar to the findings of Andrews-Casal et al. (1998), parents in this study stressed that knowing the risks would not have affected their decision to have children. However, many parents stated that they would have liked the opportunity to seek further information and prepare in advance for the possibility of their child being born with a cleft. Most patients with CL/P are discharged from cleft services around the age of eighteen years. Cleft teams should endeavour to provide all patients who are leaving the service with appropriate information and possible routes to access cleft services in the future. This should include access to genetic counselling to equip young adults with the necessary information and support they may need when it comes to making decisions about starting their own family. This could be offered as part of the transition into adult healthcare. To assist with this, clinicians may require the support of further research in order to develop relevant materials. Patients who have already left the cleft service may be more difficult to contact. General awareness-raising within the public domain and appropriate signposting from non-specialist healthcare professionals may be useful.

All parents had contact with non-specialist HPs both before and during pregnancy. In many cases, the issue of CL/P had not been raised at all. In a number of cases, the parents had raised the issue with HPs themselves, but most had been met with unhelpful responses. While many parents had received no information whatsoever, some had received misinformation about the aetiology of CL/P and the likelihood of their children being affected. It is clear from this study and many others in this field (Knapke et al., 2010; McCorkell et al., 2012; Nelson et al., 2012) that non-specialist HPs often lack the knowledge and skills required to support parents

appropriately, and basic training should therefore be recommended. HPs such as primary care physicians and midwives are typically the first point of contact for those wanting to start a family. As such, these HPs are in an ideal position to provide initial information and signpost the families to an appropriate specialist, such as a genetic counsellor. Cleft lip has recently been added to the list of anomalies that are screened for at the 20-week antenatal scan in the UK. While the sonographer's time with a patient is limited, having a prior awareness that the parent has a diagnosis of CL/P themselves and the ability to start a helpful dialogue with the parent would be a useful way to raise the issue and signpost parents appropriately. Of equal importance is the possibility that patients who are under stress or in shock may perceive a lack of support from HPs irrespective of the HP's actual competence and knowledge. This emphasises the importance of good communication skills and empathy in HPs, who may need to be aware of a patient's reduced capacity to assimilate information during stressful times.

On a wider scale, research investigating the causes of CL/P and the factors which affect heritability is of vital importance. CL/P aetiology is undoubtedly complex and difficult to investigate. Nonetheless, the aetiology of CL/P is a concern which has been identified in many previous studies and in some cases has been known to cause considerable distress to parents (Nelson et al., 2009a; Williams et al., 2011). This is an issue which affects multiple generations of an entire family, and further work is therefore urgently needed in order to offer information which is as clear and accurate as possible.

#### *Reactions to child's diagnosis*

In this study, parents of children who were born with CL/P spoke about their reactions to their child's diagnosis, while parents whose children had been born without CL/P speculated on how they may have felt had their child received a diagnosis. The findings highlight parents' potential need for support whether CL/P is present in their child or not.

A number of parents identified a variety of distressing reactions. Some of these reactions paralleled those identified in other studies by parents who do not have CL/P themselves, such as shock, guilt and concern for their child's future (Nelson et al., 2011). However, these reactions were often expressed differently by the



parents in this study. Although it has been suggested in previous research that parents who have a cleft themselves will experience more feelings of guilt or responsibility than parents without a cleft (O'Hanlon et al., 2012), the results of the present study do not enable a comparison of these different groups of parents. Nonetheless, parents in this study did comment that they would feel responsible, since the cause of their child's cleft would be 'less ambiguous' than if there wasn't a family history of CL/P. Many parents also reported experiencing feelings of sadness for their child, since unlike parents with no experience of CL/P, they knew what their child was likely to go through when growing up with a cleft, and how long the treatment journey would be. Other previous research has shown that parents who make an internal (self-blaming) attribution in relation to the cause of their child's cleft are likely to experience higher levels of anxiety and stress as a result (Nelson et al., 2009a). One finding not identified in previous literature suggested that prior to having children, some parents believed their experience of CL/P had come to an end. Having a child with a cleft subsequently meant that they had to go through the process a second time, which had caused significant distress in a small number of cases. Taken together, these findings suggest that parents with CL/P may require support to cope with feelings of guilt and sadness which arise from having a child with CL/P.

Despite this, several parents in this study commented that although they would experience feelings of responsibility for their child's cleft, they did not believe that a diagnosis of CL/P would be a significant problem for their child. This view was often based on the belief that they had adjusted well to their own condition, at a time when treatment and support may have been of a poorer standard. Subsequently, not only would their child also adjust well to CL/P, but their child's experiences of treatment would be much improved in comparison to their own. A number of parents also stated that relative to other conditions, CL/P was a minor affliction that would not significantly jeopardise their child's outcomes. Making 'downward' comparisons such as this has been identified previously in similar studies (Stock and Rumsey, in press; Nelson et al., 2011; O'Hanlon et al., 2012), and appears to be a common and positive coping strategy in parents of children with health conditions.

As a whole, these findings indicate that those adults who are less well-adjusted to their own condition may be more vulnerable to distress when considering starting their own family.

### *Factors affecting parental adjustment*

A number of additional factors which may increase or reduce the risk of psychosocial distress were identified in this study. Parents who had children with CL/P took comfort in the belief that their child's cleft was 'less severe' than their own; equally parents who perceived their child's cleft to be 'worse' than their own experienced more distress as a result. Parents whose previous children had been born without cleft, or whose 20-week scan failed to detect their child's cleft, found their child's diagnosis to subsequently be a 'bigger shock', due to having been previously reassured that they had 'managed to avoid it'. These reports indicate the need to provide parents with consistent and reliable information, and realistic expectations about what to expect for their child. This is true for all parents and patients affected by CL/P; for example, having unrealistic expectations of potential treatment outcomes can lead to distress (Cadogan and Bennum, 2011). However, it is possible that considering their first-hand experience, parents may feel they know more about cleft treatment than HPs; a potential challenge for clinicians.

For all parents, other people's unhelpful reactions and lack of understanding of CL/P had the potential to impact on adjustment. A number of parents of children with CL/P felt that family members had blamed them for their child's condition; in some cases this appeared to result in family tension for some time after their child's diagnosis. The perception of being held responsible for their child's cleft by others appeared to provoke feelings of shame in some parents. Parents' partners also had to adjust to the news that their child had a cleft. Some parents commented that although their partner had been very supportive, receiving their child's diagnosis of CL/P had caused a rift in their relationship. Unlike other new parents with no experience of CL/P, a parent with CL/P themselves has first-hand experience and unique feelings about what it means to grow up with a cleft. Discussing such feelings with their partner may be difficult, and their partner may struggle to understand. Supporting couples to communicate these feelings may be a useful way of strengthening the parents' relationship and helping them to cope with their child's diagnosis together. Within the general literature, a lack of social support is known to jeopardise wellbeing and adjustment; equally, a strong social support network can facilitate coping and encourage positive adjustment (Cohen and McKay, 1984; Baker et al., 2009). Within CL/P research, the views and support needs of partners and other family members are often neglected (Stock

and Rumsey, in press). Further exploration of the family unit as a whole may highlight a number of untapped support needs, as well as opportunities to promote helpful social support systems.

Parents in this study also commented that unwanted reactions from members of the public, such as staring and comments, seemed to occur more frequently when both parent and child had a cleft. Although the perception of social stigma is often reported in studies of this type (e.g. Carroll and Shute, 2005; Strauss et al., 2007; Hearst et al., 2008), it is a unique finding that this may be perceived to occur more often, or in a slightly different way to those parents who do not have CL/P themselves. In a more general sense, these perceptions could be influenced by parents' level of vulnerability and/or feelings of guilt. The present study is not able to compare the different perceptions of these sub-groups of parents. However, an awareness of this issue in general, and the ability to offer parents useful coping strategies, may reduce the impact of unwanted social attention (Stock et al., 2013). Raising awareness of CL/P among the general public also remains a priority.

As discussed above, non-specialist HPs can lack knowledge and skills when engaging with parents prior to, and following their child's diagnosis. This was also the case after parents' children had been born, when insensitivity from HPs was often reported. HPs tended to rely on parents to 'fill in the gaps' in HPs' knowledge, since parents had experienced cleft treatment themselves, and neglect the fact that parents may still require support of their own. Similarly, while some parents in this study were happy to speak to other families affected by cleft, they felt that they were often imparting information that should have been the responsibility of the HP. Previous research among families affected by CL/P and other conditions has demonstrated that providing parents with opportunities to engage in peer support can be of significant benefit to both parties (Stock and Rumsey, in press; Law et al., 2002; Kerr and McIntosh, 2000). However, too much reliance on this approach by HPs may put unwanted pressure on parents. Organisations such as the Cleft Lip and Palate Association (CLAPA) in the UK offer peer support on a formal and informal basis, provided by trained volunteers with personal experience of CL/P. Such an approach could reduce the burden on parents as well as HPs, and ensure that the quality of the support provided is consistent and monitored by others. O'Hanlon et al (2012) also identified the importance of accurate information and positive interactions with clinicians. Appropriate training should therefore be a future priority in this area.

Finally, having ‘faith’ or ‘spirituality’ was considered to be a positive source of support by a small number of participants. This has been reported previously in a number of similar studies, (Koenig et al., 2001; Meyerson, 2001) particularly when genetics play a part in the aetiology of the condition.

#### *Impact of own experiences on parenting style*

This theme revealed two distinct aspects of parents having personal experience of CL/P. For some, having a child with CL/P evoked some difficult memories. One parent felt that it was difficult not to convey their own worries to their child. This finding supports O’Hanlon et al.’s (2012) suggestion that some parents may need further support to cope with any unresolved difficulties of their own. However, many of the parents in this study believed that not only were they more relaxed and pragmatic than parents without CL/P in relation to their child’s cleft treatment, but that because of their own experiences they were the best person to offer support and advice to their child. Some parents felt that having a cleft themselves had a ‘normalising’ effect on their child, creating an environment in which CL/P was a regular part of life. O’Hanlon et al (2012) found that parents without CL/P reported feeling anxious significantly more often than parents who were born with a cleft themselves. The findings of the current paper support this and also add further insight to this observation.

A number of parents also commented on wanting to help their child to avoid some of the negative experiences they had when they were growing up, such as frequently moving schools, or not having anyone to talk to about their cleft. Some also talked about the positive experiences they’d had and wanting to make sure their child had the same opportunities, such as treatment at the best hospitals and involvement with organisations such as CLAPA. Other parents believed that they didn’t treat their child any differently because of the cleft. The variation in approaches to parenting supports O’Hanlon et al.’s (2012) finding that parents with CL/P often draw on their own experiences to cope. It also draws attention to the positive aspects of being a parent with CL/P, a view which has often been overlooked in the past.

#### *Impact of becoming a parent on own outlook*

To the authors’ knowledge, this theme has not been identified before in previous literature. Several parents reported that having their own child (whether their child had a cleft or not) had changed the way they thought

about CL/P and the way they viewed themselves. Some commented that wanting to improve the way they looked through cosmetic surgery now seemed 'trivial' because their children knew them and accepted them the way they were. In contrast, others had found that having a child had put them back in touch with health services and provoked them to seek further treatment. Prior research has suggested that adults born with CL/P can remain dissatisfied with their appearance after treatment has concluded, and desire further surgery to correct this (Sinko et al., 2005; Oosterkamp et al., 2007). Through their child, parents may learn about more modern procedures that could improve their own appearance, speech and hearing, and may opt for this if they believe that care has improved since they underwent their original cleft treatment.

A number of parents also commented that becoming a parent had provided them with a different perspective on their relationship with their own parents, and on the decisions their parents had made for them throughout their cleft treatment. Others believed that having a cleft themselves had helped them to foster a unique and close relationship with their child through shared experiences. The birth of their own child may thus provide parents with an opportunity to come to terms with their experiences of CL/P.

#### *A continuum of adjustment*

In-depth qualitative interviews with this unique group of parents have suggested that while a parental diagnosis of CL/P may impact on parents' experiences of having a child, these parents do not necessarily experience elevated levels of distress as a result. In fact, incorporating their own experiences of growing up with a cleft into their style of parenting can be considered advantageous for both the child and the parent. The important factor may not be the presence of a parental diagnosis of cleft, but where the parent falls on a continuum of adjustment in relation to their own condition. Not only does this demonstrate the importance of psychological research within this field, but it suggests that a positive cycle of adjustment can be encouraged and maintained through the provision of appropriate support.

#### *Methodological observations*

The results of this study question the utility of repeatedly attempts by researchers to compare experiences and adjustment in affected versus non-affected samples. In his 2001 paper, Eiserman comments that not only are

these comparisons rarely substantiated, but that authors often make apologies that their sample was not large enough to verify the extent of the deficiency, or that the deficiency would be found to be significant were the methodology to be appropriately refined. Furthermore, many researchers working in the field of craniofacial conditions continue to maintain a unique focus on the identification of problems and deficits, rather than on a full spectrum of experience. Although a large proportion of patients and parents adjust well to their own/their child's condition, this is rarely discussed. In addition, adjustment has been shown to fluctuate over time, according to the developmental trajectory and life events (ARC, 2009). While clinicians need to be able to identify and support patients who are having difficulties, learning from patients who adjust well to their condition is of equal importance. This is particularly important for the development of interventions, and with regard to HPs providing guidance about how to optimise outcomes for patients.

A second methodological observation relates to the heavy reliance on quantitative designs in this field. The limitations of using cross-sectional quantitative data, adopting exclusive (rather than inclusive) samples (see Feragen and Stock, in press), and the lack of consistency in the choice of outcome measures (see Rumsey and Stock, 2013) result in findings which are difficult to compare and interpret. In contrast, qualitative measures are able to “tap into important healthcare-related issues” and the “differing beliefs and behaviours” of patients, parents and HPs (Nelson et al., 2009b), highlighting issues which are directly applicable in an everyday clinical setting. In particular, flexible interview-based techniques offer a means of exploring the issues that are important to the participants themselves, rather than relying on clinicians and researchers to make difficult decisions regarding which measures may be clinically useful. Compared to other areas of healthcare, the field of craniofacial research lacks a significant body of qualitative research (Nelson et al., 2009b). If integrated appropriately using mixed methodological approaches, qualitative designs can offer complimentary insight into the experiences and support needs of patients and parents, and provide a supportive platform upon which suitable quantitative methods can be established and refined. In relation to the topic of the present paper, the use of qualitative methodology has helped to provide a new and useful perspective, where quantitative methods have previously been unsuccessful.

A number of limitations of the present study should be acknowledged. No parents with CL/P were excluded from participating in this study. However, none of those who volunteered to participate reported a history of syndromic clefting or additional related conditions (such as autism, developmental delay or learning difficulties). Where a syndrome is present, the risk of recurrence is known to be higher (NHS National Genetics Education and Development Centre, online resource). Furthermore, the presence of an additional condition may denote an added burden on the family in terms of psychosocial adjustment, supplementary treatment and longer-term outcomes (Rumsey and Stock, 2013; Feragen and Stock, 2014; Stock and Rumsey, in press). Given these additional considerations, the experiences of patients with syndromes and other related difficulties should be made a focus for future research.

Although previous research has identified gender to be an important demographic factor in younger groups of participants (Feragen et al., 2010; Pope and Snyder, 2005; Leonard et al., 1991), this study did not explore gender differences in any great depth. Further research investigating potential gender differences among adults with CL/P may be warranted.

Participants were geographically widespread across the UK and were recruited using various techniques. Nonetheless, one third of the parents in this study had at least one child born with a cleft, which is a higher frequency than might be expected according to national statistics (NHS National Genetics Education and Development Centre, online resource). The majority of participants also reported themselves to be White British, employed (full- or part-time) and in a long-term relationship. Since parents who took part in this study were self-selecting, there may be biases in the sampling. Further research with this group of parents should aim to be more representative of the population as a whole. In addition, studies involving those from Black and Ethnic Minority communities, as well as those from a broad range of socioeconomic backgrounds are required in relation to this topic and within the field more generally.

## Conclusion

The findings from this exploratory study suggest that parents' own experiences of growing up with CL/P impact on their thoughts about starting a family, their reactions to their child's diagnosis of CL/P (should this

occur), how they adjust to this in the long term and the parenting styles they subsequently adopt. Individual differences in responses to these challenges vary considerably and assumptions that the experiences of this group of parents are uniform should be avoided. The clarity and timing of information that young adults with cleft are given in relation to the chances of CL/P being passed onto their children, and the level of support that is offered to address any concerns at this point is crucial. Young adults who are thinking about starting a family should be signposted to the appropriate support services by specialist and non-specialist HPs. Consideration should also be given to how to best support young adults through this potentially difficult stage, while optimising existing strengths in those affected.

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