

The psychosocial impact of cleft lip and/or palate on unaffected siblings

Abstract

Background: Sibling relationships are among the most unique social connections, impacting significantly on psychosocial adjustment. Previous reviews in the fields of chronic illness and disability have concluded that unaffected siblings of children with long-term conditions are at risk of poorer psychological functioning as a consequence. Much research has investigated the psychosocial impact of CL/P on affected individuals and their parents, yet comparatively little is known about the impact on other close family members.

Objective: To gain a better understanding of the experience of unaffected siblings of children born with CL/P, with a view to informing service provision and support.

Design: Individual qualitative interviews conducted over the telephone/internet with five siblings and eight parents, including five sibling-parent 'pairs' from the same family.

Results: Thematic analysis identified three key themes applicable across both parent and sibling interviews: Perceptions of positive and negative impacts; Factors affecting the degree of impact; and Support for families.

Conclusions: This study provides insight into a population which is often overlooked in the context of cleft care. The analysis identified a number of sibling support and information needs, along with suggestions of how to incorporate support for siblings in practice. The findings suggest that an inclusive approach to healthcare encompassing all members of the family is essential for optimal familial adjustment.

Key words: Cleft lip and palate; psychological adjustment; sibling; family

Introduction

The birth and development of a child born with a cleft of the lip and/or the palate (CL/P) is known to impact upon family life. Both mothers and fathers of children born with CL/P have reported challenges relating to the impact of the diagnosis, caring for the child after birth, the ongoing burden of treatment and concerns about their child's future (see Nelson et al., 2012 for a review). For the child, challenges pertaining to social interaction, educational achievement, satisfaction with appearance and emotional wellbeing have also been reported (see Hunt et al., 2005 for a review). While the potential impact of CL/P on parents and the affected child is becoming clear, much less is known about the effects of CL/P on other close family members and of the support that may be required.

In both the US and the UK, more than 80% of children grow up with a sibling (US Bureau of the Census, 2005; Office of National Statistics, 2013). Sibling relationships are among the most unique, intense and long-lasting social connections one can experience (Brody, 1998; Lamb and Sutton-Smith, 2014). Through relationships with their sibling(s), children learn crucial information about their social world, including how to interact with peers and how to manage conflict (Lamb and Sutton-Smith, 2014). From the age of one, a child will spend as much time with their sibling(s) as they do with their parents (Whiteman et al., 2011). By middle childhood, their time spent with their sibling(s) will have overtaken the time they spend with parents (Whiteman et al., 2011). Sibling relationships have a large bearing on children's overall adjustment; an influence which is still observable in adulthood (Lamb and Sutton-Smith, 2014).

A number of studies have investigated the effect of a child's health condition on their siblings' adjustment. Previous reviews in the fields of chronic illness and disability have concluded that unaffected siblings of children with long-term conditions are at risk of poorer psychological functioning as a consequence (Lobato, 1983; Faux, 1991; Williams, 1997; Sharpe and Rossiter, 2002; Vermaes et al., 2012). These reviews suggest that when compared to controls, siblings of chronically ill or disabled children have higher levels of depression and anxiety, inferior cognitive development, more internalising and externalising problems and less positive self-attributes. Interactional relationships between siblings can also be impacted, including higher levels of sibling rivalry, and feelings of embarrassment, guilt and neglect (Faux, 1991). Such impacts may be influenced by a range of demographic factors, including gender, birth order and family socioeconomic status (SES; Lobato, 1983), as well as by the characteristics of the condition itself, such as the extent to which a condition is

life-threatening and the degree of impact on day-to-day family functioning (Williams et al., 1997; Sharpe and Rossiter, 2002). Differences between parent and sibling self-reports have also been observed; with parents most commonly reporting more negative effects than their child (Sharpe and Rossiter, 2002).

Although little research has specifically explored the psychological impact of a craniofacial condition on unaffected siblings, one study compared the behavioural adjustment of such siblings to that of an age- and gender-matched control group (Benson et al., 1999). Despite finding no overall differences in behavioural adjustment between groups, sibling behaviour did seem to be impacted by the visibility of the child's condition and by parental adjustment (Benson et al., 1999). Recent research in the field of CL/P has also been indicative of some negative effects on siblings of children born with a cleft. For example, a qualitative study with fathers suggested that siblings may receive less attention from family members as a result of the child's medical needs and exhibit more behavioural problems as a result of the affected child's medical needs (Stock and Rumsey, 2015). In a report by the Centre for Appearance Research (CAR; Bristol, UK), focus group data highlighted that siblings may experience distress over the child's surgical treatment and observe other people's negative reactions to the affected child's condition in a similar way to parents (Stoneman et al., 2014). Further, siblings may be excluded from hospital consultations and cleft-related events, and have less understanding of CL/P as a result (Stoneman et al., 2014). Currently, little support exists for siblings of children with CL/P. This is particularly apparent when compared to interventions offered within other healthcare settings which typically work systemically, including family-based, community-based, one-to-one and therapeutic camp interventions in the fields of paediatric cancer (Prchal and Landolt, 2009), general chronic illness (Lobato and Kao, 2005), deafness (see the National Deaf Children's Society www.ndcs.org.uk) and developmental disabilities (Shivers and Plavnick, 2015).

In contrast to these concerning findings, having a child with CL/P in the family has been reported to promote strong family bonds and positive growth among family members (Nelson et al., 2012). A study investigating sibling relationships in school-aged children with craniofacial anomalies found significantly less hostility and an increase in active helping, protective and play behaviours than in healthy children (Faux, 1991). Interestingly, reports found in the chronic illness and disability literature have also suggested that siblings may act as a buffer for stressful events and negative social interactions experienced by the child (Gass et al., 2007). In their study with fathers of children with CL/P, Stock and colleagues (2015) suggested that siblings had the

potential to be a positive influence on the affected child, in terms of providing close social support and in encouraging cognitive and speech development. Positive effects for both the sibling and the child with CL/P could therefore be observed.

The aim of the present exploratory study was to gain a qualitative understanding of the challenges and benefits associated with being an unaffected sibling of a child born with CL/P, according to the perspectives of parents and siblings. In addition, any potential interposing factors were explored. A pragmatic approach was taken in order to pose suggestions for clinical practice, community support and future research.

Method

Design

A qualitative approach was employed in the current study. In the field of chronic illness generally (Sharpe and Rossiter, 2002), and in the area of cleft and craniofacial research more specifically (Nelson, 2009), qualitative investigations are infrequent, yet desirable, in an attempt to enhance our understanding of complex issues and provide additional insight into conflicting quantitative findings. Qualitative research is also considered helpful when the research area under scrutiny is new, or is being explored from a different perspective (Morse and Richards, 2002).

Affiliations

This study was carried out as part of a large evaluation of the ‘Regional Coordinators Project’ by CAR on behalf of the Cleft Lip and Palate Association (CLAPA). Further information about this project can be found on the CLAPA website: www.clapa.com.

Recruitment

The study was reviewed and approved by the Ethics Committee for the Department of Health and Social Sciences at the University of the West of England in Bristol, UK. The study proposal was also reviewed by the Advisory Panel for the CLAPA Regional Coordinators Project, which is comprised of members of CLAPA, members of CAR, parent and patient representatives and members of related charitable organisations. The British Psychological Society Code of Ethics and Conduct (2009) was adhered to throughout the study.

Participants were self-selecting and were recruited through CLAPA. Advertisements on relevant websites, on social media sites and in newsletters were also released. All eligible participants who were available for interview within the study timeframe were invited.

Prior to their interview, participants were sent a participant information sheet describing the purpose of the study, what participation in the study would entail and their right to withdraw from the study. Written consent was obtained from each participant and an interview was subsequently scheduled. In the case of siblings, both sibling assent and parental consent was sought for those participants under the age of 16 years.

Participants

Participants were either a sibling of an individual born with CL/P ($n = 5$), or a parent of at least one child born with CL/P and at least one child born without CL/P ($n = 8$). Where possible, both sibling and parent participants were recruited from the same family, to allow for sibling-parent reports to be compared. Five sibling-parent pairs were recruited, along with three additional parents. A total of 13 participants were interviewed.

Participants were asked to provide basic demographic information prior to the interview. Siblings were aged between 11 and 31 years, with an average age of 17 years. Three siblings were male, while two were female. Parents were aged between 23 and 60 years, with an average age of 48 years. Seven mothers and one father participated. Participants identified as White British ($n = 7$), British ($n = 2$), Irish ($n = 1$) and Black Caribbean/White ($n = 2$), with one participant undisclosed. Participants were recruited from several regions across the UK, including the North East, the East and West Midlands, the South West and the South East. All participants except one parent was either employed or enrolled in full-time education, and all but one parent was either married or cohabiting. Parents reported having two or three children, with one of those children having been born with CL/P. Two siblings were older than the child with CL/P, while three were younger. Among the participating families, five children had been born with a unilateral cleft lip and palate, two had been born with a bilateral cleft lip and palate and one had been born with a cleft lip only. Although this study did not exclude participants on the basis of cleft type or the presence of an additional condition (such as a diagnosed syndrome, learning difficulty, Autistic Spectrum Disorder, Attention Deficit/Hyperactivity Disorder or developmental delay), no participants reported an incidence of any additional conditions within their family.

However, two of the parents reported their child with CL/P to have hearing difficulties, and one of the parents also reported having been born with a cleft.

Data collection and analysis

Individual, free-response interviews were conducted over the telephone ($n = 11$) or via Skype (without video; $n = 2$). The interviews were guided by an open-ended, semi-structured interview schedule, which was compiled by the authors using existing literature and experience of working with families affected by CL/P. Questions varied slightly according to the age of the child with CL/P and whether the participant being interviewed was a parent or a sibling. Interview topics included: experiences of the child's diagnosis and of communicating this diagnosis to others; experiences of the child's treatment and the impact of this on the sibling; social experiences and family relationships; areas of need or difficulty; any positive effects of CL/P on the family; and suggestions for support. Interviews were conducted until the authors were satisfied they had reached saturation in relation to the key themes.

Each interview was audio recorded and transcribed verbatim. Transcripts were then analysed separately by the first and second authors using Thematic Analysis (TA; see Braun and Clarke, 2006). 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, in line with a pragmatic framework (see Fishman, 1999). 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. Emerging themes were checked and discussed between the two coders until agreement was reached. Themes were chosen for their prevalence and/or their apparent importance (or 'keyness') in relation to the research question.

Results

The analysis identified three key themes, each with a number of supportive sub-themes. Initially, data derived from parents and siblings were analysed separately. Overall, themes were similar across all participants and few differences between parent and sibling reports were observed. Parents' and siblings' responses have therefore been merged throughout the results section (unless explicitly stated), and each theme is presented below along with the most representative quotes from the group of participants as a whole. The themes and their corresponding sub-themes are also summarised in Figure 1. Participants have been given pseudonyms to maintain confidentiality.

Theme 1: Perceptions of positive and negative impacts

This theme relates to the perceived negative and positive impacts of CL/P on unaffected siblings. Sub-themes describe the bond between siblings, sibling rivalry, sibling-reported anxiety and positive impacts on siblings.

The sibling bond

Participants reported a strong and close bond between children with CL/P and their sibling(s).

“[Sibling] definitely looks out for [child]. When they are at school they see a lot of each other...certainly far more than the other girls with sisters in the school” – Rebecca (parent).

“They are very close...possibly more so because of [the cleft] ... I would say they are a team” – Linda (parent).

The close bond was also described by participants as being a potential disadvantage for the sibling, who tended to assume responsibility.

“At school, when [child with cleft] was out around in the playground, people wouldn't really be able to understand him, so they'd ask me what he was saying...and I had to sort of take charge and explain it to them... Once I had to tell my whole class about it...sorry... (Crying)” – Jack (sibling).

Similarly, some participants reported that the child with CL/P could become over-reliant on their sibling.

“[Sibling] has been very protective of [child with cleft] from day one... It has been good for them but also to some extent [child with cleft] has become too reliant on it” – Bill (parent).

Sibling rivalry

Participants also identified a sibling rivalry, which was generally concerned with the amount of attention and time the child with CL/P received from parents and other family members in comparison to that spent with the sibling(s).

“Mum was trying to sort out [child with cleft] and I felt like I was, like no one was noticing me...because all the attention was on [child with cleft] it was just, they were almost ignoring me” – Jack (sibling).

Some parents noticed attention-seeking behaviours from siblings, such as hair-pulling and tantrums.

“[Sibling] did get quite possessive and would throw tantrums...once [sibling] pulled my hair so hard that...she pulled me virtually to the ground” – Rebecca (parent).

“Before [child with cleft] was born, a lot of the time it had just been me and [sibling], so [sibling] would get quite upset because he was losing his special time with me... He would act up a lot, try to get told off almost just for the attention” – Sarah (parent).

Some participants recalled siblings making upsetting comments.

“[Sibling] said ‘if I was ill like [child with cleft] you would have more time for me wouldn’t you Mummy?’” – Vanessa (parent).

“[Sibling] kept saying things like ‘I want to go into hospital’ and ‘I want to break my arm’ and it did really concern me at the time” – Tanya (parent).

Rivalries occurring at school were also identified.

“At school people would ask me questions about when [child with cleft] was coming back and teachers would ask me questions about how she was doing, and I used to get jealous because she was off school and I wasn’t...and everyone was asking about her and not me” – Abbie (sibling).

Sibling anxiety

Participants reported a range of concerns exhibited by siblings, which varied in their degree of severity. Many of these concerns were focused on the child with CL/P having treatment or going into hospital.

“[Sibling] wanted to know about the surgery and how it would happen, when it would happen, who would do it, and then what [child with cleft] would look like...afterwards there was blood and some stitches and [sibling] was worried [child with cleft] would be in pain” – Sarah (parent).

These anxieties generally stemmed from siblings having a lack of understanding about the treatment process.

“I was a bit shocked when [child with cleft] came back from surgery...I half expected her to be in a wheelchair or passed out or something... I just wish that someone had told me what she would look like” – Melissa (sibling).

“When I was younger I was just a bit confused...because I couldn’t really understand much of what my parents were talking about... ‘Why is he away so often? What’s going on?’” – Jack (sibling).

Anxieties that had a larger impact were linked to siblings who had to stay with friends or wider family members when their sibling was in hospital.

“[Child with cleft] went into hospital for what felt like forever... Usually Mum would give me to family or friends, and I would sort of be on my own for a while...so I had long periods of time where my mum and sister had disappeared as far as I was concerned... (Crying)” – Abbie (sibling).

Some of the more negative experiences were reported to have a longer-term impact on siblings.

[Sibling] worries now that if he gets hurt he’s going to have to have some sort of surgery himself and this really frightens him” – Sarah (parent).

“It’s still going on really...these are women in their thirties now, who I’m very proud of and they’re very successful in their lives, but this cleft lip and palate and the issues related to it is with them still” – Tanya (parent).

Positive impacts

A number of positive impacts related to being a sibling of a child with CL/P were reported. Siblings were acknowledged as being particularly caring and kind, often helping and supporting others.

“I think it has made [sibling] a very caring child...she is certainly always aware of girls in her class who may be having difficulties... I think she’s someone that everybody would say that they could confide in and get support from” - Rebecca (parent).

“It had a really positive impact on me...when [child with cleft] was born I had to connect with him...I got picked [to be a school prefect] because I was really good at helping the younger children” – Jack (sibling).

“[Sibling] has actually gone on to be a midwife...when she was applying to get onto the midwifery course she talked about [child with cleft] in the interview...how it’s not all lovely and wonderful and cuddly babies and that actually things can be quite different” – Caroline (parent).

Another positive aspect was having a non-judgemental attitude to others with a visible difference, due to the additional understanding and personal experience gained from having a family member with this type of condition.

“I have more knowledge about cleft because of it...I can explain it to my friends and things which I think is good for awareness... Also when I see other people with similar looks to [child with cleft] I would think twice about taking the mick, because they might have gone through something like [child with cleft]’s gone through” – David (sibling,).

Theme 2: Factors affecting the degree of impact

A range of factors that played a role in the degree of impact on the sibling were identified. These included birth order, age gap, gender, treatment burden, socioeconomic status, social support network, sibling involvement and parental coping.

Birth order

In comparison to younger siblings, participants stated that elder siblings were more likely to take on a protective role and provide stability for the child with a cleft.

“I think maybe because [child with cleft] has got two older siblings it’s more stable and...us being there so that if he does get bullied or needed anything we would naturally step in” – David (sibling).

In some cases, the majority of cleft treatment had taken place by the time the younger sibling was old enough to remember, and so the initial impact on the sibling appeared to be lessened.

“I don’t think [sibling] was really aware...by the time he was old enough to understand we had already gotten over the feeding problems and [child with cleft] had his palate fixed, so it was just hospital appointments twice a year” – Linda (parent).

Age gap

Where siblings were close in age, parents reported more of a struggle in the early years, predominantly because younger children were seen as more dependent.

“It is hard already having young children who are close in age and then also a baby who can’t feed normally...it takes more time and there’s much more to think about” – Vanessa (parent).

However, as they grew up siblings who were closer in age were more likely to attend the same school at the same time, and more likely to attend a cleft-related event together.

“[Child with cleft] was going to a school which his brother was already at...so I think it probably helped that when [child with cleft] came up his brother was still there” – Caroline (parent)

Gender

Siblings who were the same gender often perceived their relationship to be closer and to have a higher level of empathy with their sibling. This was thought to be due to enjoying the same activities (particularly in males) and confiding in each other (particularly in females).

“I think if [child with cleft] had been a boy...it would have been different, but we’ve got similar interests and stuff...we like arguing (laughs)...she can definitely speak to me...I’ll always be there to listen and she can trust me” – Melissa (sibling).

Participants also described the potential impact of gender on appearance-related concerns; specifically, participants felt the altered appearance due to the cleft could be more problematic for females.

“Maybe if [child with cleft] was a girl and he cared what he looked like it might affect him, because his nose is a little bit different...I guess him being a boy and being quite laid back it’s been easier” – David (sibling).

Treatment burden

Another key factor was the perceived (and actual) burden of cleft treatment. Siblings of individuals who had less need for treatment reported not noticing much of an impact on day-to-day life.

“It was kind of normal apart from sometimes she would have an appointment with an orthodontist” – Edward (sibling).

“There were additional operations, I can’t even tell you how many grommets [child with cleft] had put in...I think it impacted significantly on [sibling]” – Tanya (parent).

Social support network

The social support network, which was predominantly spoken of in relation to the wider family unit, was a key factor influencing the degree of impact on the sibling and the family as a whole. Single parents and families without a support network reported struggling with the demands of looking after their children, particularly given the additional attention a child with CL/P required.

“My family support was sporadic....so there were times when I had to leave [sibling] with friends and it wasn’t ideal...if [child with cleft] was going into hospital I would almost have to beg someone to have [sibling] for a few days...it was never consistent” – Tanya (parent).

In contrast, families for whom additional support was reliably available reported less of an impact on the sibling.

“[Sibling] stays at my mum’s quite a lot. She only lives a couple of miles up the road and it’s like his second home...we try to keep it as normal as possible for him” – Sarah (parent).

Socioeconomic status

SES impacted predominantly on the amount that siblings were able to be involved in the cleft network and treatment process, as well as on parental stress levels.

“At the time I didn’t know that you could claim for your bus fares if you were on benefits, so I used to [avoid] the fares from my house to the hospital...there were times when I felt that someone would notice all this and I lived in fear that people would take [sibling] away” – Tanya (parent).

“My husband had a difficult job which meant he couldn’t take time off easily.” – Linda (parent).

“Mum was a single parent...everything was a bit [improvised].” – Abbie (sibling).

Parental coping

Parents’ general ability to cope with the challenges related to CL/P was another factor influencing the impact on siblings. Parents reported that, especially with a new-born, stress levels were high and this could result in family conflict, with the sibling’s needs being overlooked or underestimated.

“I wasn’t getting on very well with feeding [child with cleft]...I was so tired, I was concerned about the operations...it does throw things into turmoil and you are possibly less tolerant of [the siblings]... The trouble is you...don’t think about the effect on [the siblings] really, because it’s not your main focus” – Caroline (parent).

Participants commented on siblings’ awareness of heightened parental stress.

“When [child with cleft] went in for one of her nose reconstructions, I was 15 and I went to the hospital...I suddenly realised (cries) how much my Mum had been on her own during the whole process” – Abbie (sibling).

“I think probably because I was quite tense...[sibling]’s quite sensitive and picks up on these things” – Caroline (parent).

“You have to be careful what you say around him really because he takes it all on board” – Linda (parent).

Sibling involvement

Being involved and feeling included in the process was important for all siblings.

“I was never worried about [child with cleft] going in to have the operations...I knew what they were doing and what was going on...and what the operation was going to do in the end...we were all kind of in it together” – David (sibling).

“[All the children] went along to the appointments...[the doctors] spoke to [sibling] and they were always aware that she was there...they are busy clinics, but they were still speaking to her” – Bill (parent).

Siblings tended to become more involved with later treatment, such as the bone graft operation.

“Because [sibling] was that much older when [child with cleft] had her bone graft, it was all dealt with in much more depth...[sibling] had lots of questions and she became much more involved at that point...[sibling] even took on the role of nurse!” – Rebecca (parent).

Theme 3: Support for families

Participants discussed various types of support that could have helped siblings on their journey. The type and level of support requested often varied according to the direct recipient of the support and the sibling’s age.

Parents’ and siblings’ support needs

Participants reported that siblings were often left out of the healthcare system.

“It was obvious that the health professionals’ priority was [child with cleft] ... I don’t think they really made a fuss or paid much attention to [sibling]she just played in the corner on her own and let the meetings take place” – Rebecca (parent).

“...so Mum and [child with cleft] would ask all the questions that they wanted answers to...but I just stayed very quiet, just sat back because...I didn’t feel like it was really my place to ask questions” – Abbie (sibling).

Participants commented how a greater involvement in the treatment process and a better understanding of CL/P would have been beneficial for siblings.

“...just to be reassured that all of these are normal feelings...understanding that surgeons are very skilled and it’s routine for them...maybe going into the processes of it...that kind of thing would be so helpful, giving some control back to the sibling, because you do get forgotten” - Abbie (sibling).

“I asked my mum some questions but a Doctor would probably be able to describe the detail of it a bit more” – Jack (sibling).

“If he’d had more knowledge and understood it...he could have coped with anyone saying ‘your brother looks funny’ or whatever” – Linda (parent).

Parents also expressed a need for support to explain CL/P and its treatment to the child’s sibling(s).

“It was quite hard sometimes answering some of the questions, because I didn’t even really know the answers myself... I also never really knew how much of it [sibling] was taking in or how to explain it in a way they would understand” – Sarah (parent).

It was uncommon for siblings to have met other siblings or children affected by CL/P, and to have attended any cleft-related events. Participants believed this level of involvement could have many potential advantages for siblings.

“I did go to one family day...it helped me get a better understanding of what it was actually like for all these children, because before then I’d only really seen my brother....I remember meeting one or two [other siblings] ...they were pretty much like me in a way, we had quite a few things in common...it felt good to talk to someone else who understood about it...to see what their experiences are and see what helps them when things are going a bit, like, when it’s difficult” – Jack (sibling).

Some participants also commented about the heritability of CL/P, and the potential effect of this on siblings.

“[Sibling] has asked me and his dad that if we had any other children whether they would have a cleft...we’ve said we can go to the geneticist...because he has obviously got concerns...in time his concerns might also be that he would have children with cleft...he might want to find out things like that as he gets older” – Sarah (parent).

Mode of support

Participants had a number of suggestions for how to improve support for siblings. Many of these suggestions were aimed at helping the sibling understand the treatment process and become more involved where possible.

“Possibly a book or a comic or a DVD or something to show you the processes” – Abbie (sibling).

“Before and after surgery pictures and stuff were shown to us, but not to [sibling], it would have been nice to have something to show him” – Sarah (parent).

Some participants commented that improving some of the hospital facilities or the support available to the family at key times during the treatment pathway would have increased the sibling’s involvement and reduced familial anxiety.

“Just a bit of security for [sibling], if I could have brought her with me, or if they had a carer there, or if she could have gone to the crèche...even if there was a ‘buddy’ system with the other families...if [sibling] could have been a part of it I think that could have reduced my anxiety and could have helped her to understand the hospital environment...she could also have been there when the doctors came round to see [child with cleft]” – Tanya (parent).

Sometimes the lack of inclusion was due to the sibling being at school when appointments or events were held.

“A lot of these things were during school time, so [sibling] didn’t come to [events] and never really had the opportunity to ask health professionals questions” - Rebecca (parent).

Participants frequently commented that existing events and activities didn’t seem to be tailored to siblings.

“The people there are not really anywhere near my age...also the events are sort for the children with clefts and because I haven’t got one myself it’s not really the place to be, if you get what I mean...if it was with the sisters and brothers and stuff I think it would have been quite useful” - Melissa (sibling).

Participants suggested that events that were inclusive of siblings, or specifically for siblings would be valuable.

“A sibling group would be a good idea, like when the children with clefts go on a weekend away and do team building and stuff...a siblings-only one of those that would have been good” - Melissa (sibling).

“If you had a workshop where we can play games and they explained about cleft too, maybe you could have one in the holidays... Or maybe if you had a youth club for siblings...so they could make friends and ask questions and things” – Jack (sibling).

Although a number of benefits to online or remote support were acknowledged, on the whole face-to-face support was preferred.

“Both [online and face-to-face] would be good really...I’ve got my phone and my laptop so asking questions online would be easy...calling someone [over Skype] would also be easy” - David (sibling).

“I think in person is more personable really and it just helps you a bit more because you can see the person instead of being online, or a phone call” – Melissa (sibling).

Discussion

To the authors' knowledge, this is the first study to explore the psychosocial impact of CL/P on unaffected siblings from a qualitative perspective. The findings derived from this study suggest a range of potential positive and negative impacts on siblings, as well as a variety of interposing factors. This study also highlighted a number of possible ways to improve support for siblings of children with CL/P in both clinical practice and within the CL/P community as a whole.

Sibling relationships

A strong bond between children with CL/P and their siblings was described. Siblings were reported to adopt a protective role, to spend large amounts of time with the child and to share friendships with the child. While close sibling relationships are frequently observed among children who are not affected by a health condition (Lamb and Sutton-Smith, 2014), it is possible that the experience of CL/P can intensify this bond, as perceived by participants in this study. Nelson and colleagues (2012) previously discussed the potential for CL/P to promote strong bonds between family members, while a study by Faux (1991) detected an increase in active helping, protective and play behaviours in sibling relationships among children with craniofacial anomalies when compared to healthy controls. In a previous study with fathers of children with CL/P, participants perceived siblings to have a positive influence on the affected child, both in terms of social support and in encouraging their cognitive and speech development (Stock and Rumsey, 2015). Elsewhere, studies have suggested that close relationships can act as a protective buffer against negative social experiences in children with CL/P and in general (Erdley et al., 2001; Feragen et al., 2010), a finding which is echoed in the chronic illness and disability literature (Gass et al., 2007). In contrast, this closeness was also perceived by participants to have a potentially negative effect on both the sibling and the child with CL/P. Some siblings appeared to assume a high level of responsibility for the affected child's wellbeing, while children with CL/P were reported to occasionally rely too much on their siblings for support. Some siblings were reported to have engaged in arguments or physical fights in order to protect the child. These findings seemed to be exacerbated if the sibling was older, if there was a smaller age gap and if siblings were of the same gender as the child with CL/P. An overprotective parenting style has been found to increase social anxiety in children with chronic illness and to correlate with a reduced quality of life in both child and parent; a finding which is often mediated by

perceived child vulnerability (Anthony et al., 2003; Hullmann et al., 2010; Pinquart, 2013). It could be that siblings can assume a similar protective role, and thus comparable effects are observed.

As well as a strong bond, rivalries between siblings were reported by participants. This competitiveness was generally concerned with the amount of attention and time the child with CL/P received from parents and other family members, in comparison to that given to the sibling. Attention-seeking and aggressive behaviours were often observed by parents. In some cases siblings had commented that they wanted to become ill or to go into hospital in order to receive the same attention as the child with CL/P. Such rivalries were reported to be more common if the sibling was older, due to siblings having received more attention before the child with CL/P was born. This rivalry also extended to the school environment, where participants felt the child with CL/P was the recipient of more attention from peers and teachers. Previous studies have suggested an increase in externalising behaviours among siblings of children with chronic illness (Lahteenmaki et al., 2004; Vermaes et al., 2012), including those affected by CL/P (Stock and Rumsey, 2015). Rivalries are common between all siblings in the general population (Lamb and Sutton-Smith, 2014). However, it may be that these rivalries are intensified if one of the children has a chronic illness, as suggested by previous literature (Faux, 1991). Longitudinal research is recommended, with a view to better understanding development, expression and impacts of complex sibling relationships on the psychosocial adjustment of both the sibling and the child with CL/P.

Support for siblings and parents in clinical practice

Participants' reports indicated that all siblings had experienced varying degrees of anxiety. This echoes previous research which has described an increase in anxiety, social withdrawal and depressive symptoms among siblings of children with chronic illness compared to controls (Sharpe and Rossiter, 2002; Lahteenmaki et al., 2004). In the present study, much of the reported anxiety seemed to stem from siblings' lack of understanding of CL/P and its treatment. Contrastingly, those siblings who understood the condition and felt involved in the family's efforts to cope with the impacts reported much less anxiety about the child's treatment. The perceived impact of this seemed to vary according to the age of the sibling, since levels of understanding increased with age. Parents reported needing support to explain CL/P and the treatment process to the affected child's siblings. Concurrently, all participants felt siblings were often excluded from the provision of care, with few opportunities to ask cleft professionals their own questions. Issues raised included the need to be reassured

about the impacts of treatment on their brother or sister, to understand more about what would happen during treatment and the likely outcomes, and support in developing strategies to explain CL/P to peers. As well as contact with health professionals, participants suggested that materials, such as a story book or a DVD, may enhance siblings' understanding of CL/P and its treatment, and facilitate important conversations between family members. It was also suggested that 'before and after' surgery photographs of other children could be useful when explaining treatment to siblings, and could reduce the shock experienced by some siblings in this study following changes to the child's appearance after surgery. In the UK, cleft teams produce leaflets and other materials to explain CL/P to affected children and their parents. It may thus be beneficial to adapt some of these materials with siblings in mind. In other areas of healthcare, one-to-one and family-based interventions have already been advocated for improving siblings' knowledge of the condition and reducing emotional and behavioural difficulties (Lobato, 1983; Sharpe and Rossiter, 2002; Prchal and Landolt, 2009). Participants also commented that having a hospital environment which was more accommodating of the family as a whole could have prevented long periods of separation and increased the sibling's sense of involvement in the process.

Support for siblings and parents in the community

Participants reported that siblings were also often overlooked in terms of cleft-related events. While some participants described the potential benefits of attending events targeted at families, others commented that existing events were not always appropriate for siblings, due to the timing of events and/or the age range for which the activities were designed. These findings have relevance for organisers of existing events and suggest that new events and activities designed specifically for siblings may be advantageous. Specifically, participants advocated weekend trips, workshops, youth clubs and online support, in which brothers and sisters could meet other siblings of children affected by CL/P and have the opportunity to ask questions in a fun and comfortable environment. In the related fields of paediatric cancer (Prchal and Landolt, 2009), general chronic illness (Lobato and Kao, 2005), deafness (see the National Deaf Children's Society www.ndcs.org.uk) and developmental disabilities (Shivers and Plavnick, 2015), events for siblings are much more commonplace. These include regular sibling groups and one-off therapeutic camps. In the UK, peer activities for those affected by CL/P (see www.clapa.com) and other visible conditions such as burn scarring (see <http://www.britishburnassociation.org>) already exist. It may therefore be beneficial to consider extending these opportunities to siblings as well.

Some siblings reported distress in relation to an enforced separation from their family during the affected child's surgery. In some cases this separation appeared to have had long-term effects. This anxiety was aggravated if the family reported lower levels of social support and were unable to offer siblings a familiar or consistent place to stay while the affected child was receiving treatment. Concurrently, siblings of children who required more frequent episodes of treatment reported higher levels of disruption. Participants suggested having the option of a 'buddy' system with other families, to provide them with additional practical support, such as childcare or shared transport, around the time of key events such as operations. Again, there may be an opportunity for representative organisations, in collaboration with cleft teams, to offer this level of support to families who require it.

Fostering positive outcomes

All participants identified both negative and positive impacts of CL/P on unaffected siblings. Positive impacts have not been explored in this context previously and this study therefore offers some interesting insight. Specifically, brothers and sisters were described as being particularly caring and kind, with some later opting for careers in the care industry. Siblings were also described as having a non-judgemental attitude towards people with other forms of visible difference or health conditions as a result of their family's personal experience. Similar effects have been observed among siblings in the wider literature, with siblings reporting positive consequences such as early signs of maturity and greater levels of empathy and compassion (Sargent et al., 1995; Sloper, 2000). Positive impacts and personal growth have also been reported within the field of CL/P among affected individuals and family members (Nelson et al., 2012; Stock et al., in press 2015). Such findings suggest that with the right support, the impact of CL/P can be turned into a strengthening experience for all members of the family.

Potential interposing factors

In line with previous research with siblings of children with health conditions, this study identified a number of potential interposing factors on sibling adjustment. These included gender, birth order and family socioeconomic status (SES; Lobato, 1983), as well as treatment burden (Williams et al., 1997; Sharpe and Rossiter, 2002) and degree of parental adjustment (Benson et al., 1999). The idea that appearance concerns may be more prevalent and problematic in girls has been debated in previous qualitative (Stock et al., in press)

and quantitative (see Smolak, 2004) research, although the relationship seems far from straightforward (see Fawcner, 2013). In relation to treatment burden, a greater number of operations and appointments are typically associated with higher demands on parents, which is likely to impact further on the functioning of the broader family (Kramer et al., 2007; 2009). Family SES is known to impact upon many areas of life, and may interact with the challenges associated with CL/P, such as practical difficulties in attending frequent hospital appointments (Stock et al., in press), having to take time off work to attend appointments (Stock and Rumsey, 2015) and being unable to afford consistent childcare, as indicated in the present study. In addition, how parents adjust to the challenges is likely to have a significant impact on the child's own psychological development (Nelson et al., 2012). Unfortunately, the direction of the effects of birth order and gender were not clear from previous research, and thus more detailed comparisons with the present findings are not possible.

The present study also highlighted some additional factors which may affect the impact of CL/P on siblings, including the age gap between the affected child and the sibling, the effectiveness of the social support network within and beyond the family and the level of sibling involvement in dealing with the challenges of cleft. In relation to age gap, parents had found it more difficult to manage the challenges of having a child with CL/P if the child and their sibling(s) were close in age. This was largely due to the sibling being more dependent on parents, in addition to parents having to manage the CL/P, its diagnosis and its treatment. Relatedly, the ability of parents to cope with the consequences and challenges of their child's condition, as well as the amount of perceived social support available to families, affected the emotional wellbeing of siblings and the degree to which siblings could be involved in the treatment process. These reports suggest that providing psychological support to parents throughout their child's treatment journey can have an indirect positive influence on the wellbeing of the family as a whole. Similar findings have been reported in a recent study with fathers of children with CL/P (Stock and Rumsey, 2015); another previously neglected patient group. An awareness of the needs of the entire family, and taking an inclusive approach to healthcare wherever possible, is therefore recommended (also see Phillips and Rumsey, 2008; Burke and Montgomery, 2001).

Another potential interposing factor was identified in a previous study investigating the possible effects of craniofacial conditions on unaffected siblings from a quantitative perspective (Benson et al., 1999). Although no overall differences between siblings of children with craniofacial conditions and controls were observed, the study did suggest that the visibility of the child's condition could play a role (Benson et al., 1999). The present

study was not in a position to discuss the impact of visibility, since all participants were siblings or parents of children with visible clefts (i.e. cleft lip only, cleft lip and palate). However, subjective perceptions of visibility have clearly been shown to more accurately predict psychological adjustment when compared to more objective measures of visibility, and in contrast to parent/teacher proxy reports (Appearance Research Collaboration, 2009). This factor may thus require further investigation in relation to sibling adjustment.

Methodological considerations

Although themes were common across both parent and sibling reports, there was an anecdotal indication that parents may underestimate the impact of CL/P on the sibling when compared to the siblings' own reports. Interestingly, this finding is in contrast with previous reports in the chronic illness literature, which have found parents to report more negative effects on siblings (Sharpe and Rossiter, 2002). However, in their investigations of the impact of CL/P on family functioning, Kramer and colleagues (2007, 2009) detected a low impact on siblings, as reported by parents. Several parents in the current study commented that throughout their journey, their focus had been on the child with CL/P, and that they had not really considered the potential impact on the sibling(s) until this study was advertised. Sibling participants had been sensitive to this, frequently reporting that they felt 'ignored' by the family or 'left out'. Within and outside of the field of CL/P, several studies have highlighted differences between parent and child reports, and thus the value of capturing multiple perspectives (Goodman, 2001; Chang and Yeh, 2005; Feragen et al., in press 2015). In addition, qualitative research such as the present study can offer further insight into the findings derived from quantitative research, particularly if these quantitative findings are conflicting. This complementary line of enquiry also has the ability to explore patient groups who have previously been overlooked, and has the potential to inform future quantitative studies (Nelson, 2009).

This study identified a number of positive impacts on siblings of children born with CL/P. These findings, together with previous research, emphasise the need to measure patient-reported protective factors, resilience and personal growth, in addition to difficulties and negative impacts. Such an approach would allow for a balanced view of the experience of CL/P and support care providers to foster patients' strengths, as well as to address challenges.

A number of limitations of the present study must be acknowledged. First, participants were self-selecting and recruited through a CL/P UK charity (CLAPA), and therefore the sample is unlikely to be wholly representative. This limitation is also strengthened by the relative lack of participants from a minority ethnic community, as well as those families affected by additional conditions or syndromes (see Feragen and Stock, 2014). Nonetheless, both mothers and fathers participated in this study, as did male and female siblings of varying ages. Geographical representation from around the UK was also reasonably widespread. Future research should endeavour to investigate the needs of these often excluded groups, as well as other potentially vulnerable subgroups.

For this study, parents and siblings were recruited from the same family wherever possible, so that multiple perspectives could be collected, and so that parent and sibling reports could be compared. While this contributed an interesting dimension to the study, it also may have had a negative impact on recruitment. Although appropriate for a qualitative study of this type, the sample is smaller than the authors would have liked. Nonetheless, the authors were satisfied that saturation had been reached in relation to the key themes identified in this study, and advocate the use of these findings to support future qualitative and quantitative research in this area.

The research design employed in this study included the use of Skype (without video). Skype allows for free telephone calls to be made across the UK, eliminating call and travel costs, and is a promising cost-effective tool for qualitative research (Carter, 2011; Janghorban et al., 2014). However, using Skype in the current study proved to be more challenging than using the telephone as a method of data collection. Initially, extra communication between researcher and participant was required in order to set up the interview session. Additional complications also arose during the interview, with the connection being lost and the interview losing its flow. Following data collection, transcription time was increased due to sound interference. In the present study, utilising Skype (without video) did therefore not appear to provide many benefits over and above using the telephone. Video was not included in this study so as to ensure comparability between Skype calls and calls conducted over the telephone. However, the option of including video in a call may be valuable for future research, since face-to-face interviews can provide additional visual cues in a way that is relatively comparable to interviews conducted in person (Janghorban et al., 2014).

Finally, it is important to note that although potential interposing factors, such as birth order, were explored, these were only discussed in relation to participants' qualitative data. It is not considered possible, nor was it the intention of the authors, to compare (for example), the adjustment of older versus younger siblings quantitatively in this study. Nonetheless, this exploratory study provides insight into factors which could be measured quantitatively in the future.

Conclusions

This study provides unique insight into a population that has been previously overlooked in both CL/P research and practice. In-depth individual interviews with parents and siblings identified a number of support needs, along with suggestions of how to incorporate support for siblings in clinical practice. The findings highlight the benefits of utilising qualitative approaches to better understand the experiences and support needs of previously neglected patient groups, as well as the importance of capturing multiple perspectives. The findings also suggest that an inclusive approach to healthcare, encompassing all members of the family, is desirable in order to optimise outcomes for all. As well as 'normalising' the process for siblings and reducing anxiety, being involved in the treatment process may empower them to learn about CL/P, to feel included and to report positive consequences of growing up in a family affected by cleft. Those concerned in the delivery of CL/P services may be interested in interventions currently delivered in other health fields, with the aim of optimising the psychosocial adjustment of unaffected siblings.

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