Parents' experiences of postnatal diagnosis and care following the birth of a child with cleft lip and/or palate

Abstract

Background: Receiving the news that their child has a health condition, such as a cleft lip and/or palate (CL/P), can have a considerable psychological impact on parents. Yet previous research has highlighted parental dissatisfaction and service-related issues.

Aims: To investigate the experiences of parents of children born with CL/P in relation to postnatal diagnosis and neonatal care.

Method: Data were collected from 470 parents using an online mixed-methods survey.

Findings: The majority of participants received their diagnosis and postnatal care from a midwife. 61% were satisfied with their overall diagnostic experience. However, participants also perceived a lack of sensitivity, knowledge and empathy from hospital staff. Further issues were raised regarding the implications of a 'delayed' diagnosis including feeding difficulties.

Conclusion: Better training and support for midwives is recommended to address the challenges associated with the postnatal diagnosis and neonatal care of children with CL/P.

Key words

Cleft lip and palate; parent experience; postnatal diagnosis; neonatal care; feeding; mixed-methods

Key points

Why is this research needed?

The accurate, timely, and sensitive delivery of a diagnosis of cleft lip and/or palate is vital for the health of the child and the psychological adjustment of the family. Yet previous research has been indicative of parental dissatisfaction and service-related issues.

What are the key findings?

A considerable proportion of participants perceived a lack of sensitivity, knowledge and empathy from non-specialist health-professionals, and 39% of participants expressed dissatisfaction with their overall diagnostic experience. A perceived lack of knowledge among non-specialist health professionals was associated with parental psychological distress, feeding difficulties, and a delay in the detection of cleft palate.

How should the findings be used to influence policy/ practice/ research/ education?

The findings indicate a need for further work to understand how to better support non-specialist health professionals to provide a timely diagnosis and to support new parents, in addition to improving links between local hospitals and specialist CL/P organisations.

Introduction & background

A cleft lip and/or palate (CL/P) is one of the most common congenital conditions in the world and affects one in every 600-700 infants each year in the UK (Cleft Registry & Audit Network, 2017). It is caused by a complex interplay of genetic and environmental factors (Dixon et al., 2011), and can affect the lip, the palate, or both. In some cases, a cleft palate may be associated with additional conditions or syndromes, such as Pierre Robin Sequence, and 22Q11.2 Deletion Syndrome. While the primary surgery to repair the cleft is usually performed during the child's first year of life, CL/P can often lead to issues with feeding, speech, teeth, and hearing. As such, babies born with CL/P will require ongoing treatment to address the functional and appearance-related consequences in accordance with the 20-year UK care pathway (NHS England, 2013). The cleft and its associated treatment may also impact on several domains of the child's psychological and social functioning throughout the lifespan (Stock & Feragen, 2016).

In countries with a high level of resource, cleft lip is usually identified antenatally. In the UK, it became a requirement to routinely assess the foetal face during the 18-21 week anomaly scan in 2010 (Public Health England, 2013). This has since led to a 75% national standard detection rate for cleft lip. However, a cleft palate is rarely detected during routine antenatal screening and therefore almost all cases of cleft palate are diagnosed following the birth. In the case of isolated cleft palate (where the lip is not affected), the neonatal examination becomes crucial for a timely and accurate diagnosis, where a diagnosis outside of the first 24 hours following birth is considered to be 'delayed' (Bannister, 2008). Following the publication of evidence to suggest a significant and unacceptable delay in the detection of cleft palate, and the adverse impact of this delay on families (Habel et al., 2006; Cleft Registry and Audit Network,

2012), new guidelines were released by the Royal College of Paediatrics and Child Health (2014). These guidelines incorporate visual rather than digital assessment of the palate, in efforts to improve the routine new-born palate examination. Following diagnosis of cleft lip and/or palate, the national standard is for new-borns to be seen within 24 hours of referral by a cleft Clinical Nurse Specialist for a detailed feeding assessment and ongoing management.

For the parents and the wider family, the discovery that their child has CL/P can be a shocking and distressing experience, irrespective of whether the diagnosis is made antenatally or postnatally (Nelson, Glenny, Kirk & Caress, 2012). The manner in which the diagnosis is delivered can also have an impact on parents' wellbeing and their ability to adjust to their child's condition in the longer-term (Maes et al., 1998; Matthew et al., 1998; Chuacharoen et al., 2009; Vanz & Ribeiro, 2011). Yet, previous work in this field has suggested that parents often report a suboptimal experience with non-specialist health-professionals, including sonographers, midwives, health visitors, and GPs (Johansson & Rigberg, 2004; Knape, Bender, Prows, Schultz & Saal, 2010; McCorkell, McCarron, Blair & Coates, 2012; Stock & Rumsey, 2015), and a lack of reliable information and support (Searle, Ryan & Waylen, 2016; Young, O'Riordan, Goldstein & Robin, 2001). Further, infants born with a cleft palate often struggle to feed; a challenge which can cause additional distress in the parents (Lindberg & Berglund, 2013; Searle et al., 2016).

Aims

Given the critical importance of the diagnosis and the subsequent care the family receives, the objective of the present study was to examine the experiences of a large sample of parents in relation to their child's postnatal diagnosis and neonatal care. In

doing so, this study hopes to encourage a better understanding of parents' experiences and to offer pragmatic suggestions for improving practice.

Method

Design

A mixed-methods survey was designed and distributed by the Cleft Lip and Palate Association (CLAPA) using the online survey platform, SurveyMonkey. The survey was designed with comments and feedback from parents and health professionals from the CLAPA community. The full survey consisted of nine demographic questions (including the parent's age at the time of diagnosis, and their child's cleft type), 27 quantitative questions (including who provided the diagnosis, and how satisfied parents were with their diagnostic experience), and five qualitative questions (including the ways in which the amount, timing and content of diagnostic information could be improved). A full list of survey questions is provided in Table 1.

Procedure

Institutional approval to analyse the data was provided by *(university),* and a data sharing agreement between the university and CLAPA was signed and agreed by all parties. The study was conducted in line with ethical guidelines provided by the British Psychological Society such that participants were made aware that their involvement was voluntary, that data would remain confidential, and that they could withdraw from the study at any time up until the completion of the analysis. The survey was advertised to parents of children born with CL/P on the CLAPA website, e-newsletters and social media between October 2016 and January 2017. A total of 1,044 eligible survey responses were collected. Of these, 470 respondents reported receiving a postnatal

diagnosis. This manuscript outlines the quantitative and qualitative data provided by these parents, specifically on the topics of postnatal diagnosis and care.

Data analysis

Quantitative data was analysed by (*author*) using descriptive statistics. Since not every respondent answered all the questions on the survey, percentages are reported along with the number of responses obtained. Qualitative data was analysed by (*author*) using inductive content analysis; a method which is appropriate for summarising a large body of qualitative data (Neuendorf, 2017). (*Author*) read through the dataset several times and derived common themes using an iterative process. (*Author*) also coded a proportion of the data to assess reliability. Codes were then compared and both researchers discussed any discrepancies until full agreement was reached. Finally, frequency counts were calculated and exemplar quotes were selected to illustrate each category.

Results

Participant characteristics

Table 2 presents demographic information on the 470 parents who participated in this study. Figure 1 provides a visual presentation of year of diagnosis (note that year of diagnosis data is missing for 20 parents).

Quantitative Findings – all respondents

Overall, most respondents reported being either very satisfied (15%, 72/466), satisfied (20%, 93/466), or neutral (26%, 121/466) about their diagnostic experience. However,

19% (89/466) reported being dissatisfied and 20% (91/466) reported being very dissatisfied with their experience.

The majority of respondents reported that the news of their child's diagnosis had been presented very sensitively (32%, 152/466) or somewhat sensitively (38%, 178/466). However, 16% of respondents (73/466) felt the diagnosis had not been delivered sensitively, while 11% (52/466) felt the diagnosis had been delivered very insensitively. Only 50% (177/356) felt the hospital staff were knowledgeable about CL/P and just 59% (208/356) felt that the hospital staff were sympathetic about what the diagnosis could mean for feeding. Furthermore, only 67% of respondents (236/356) reported that hospital staff had used the correct term for 'cleft lip and/or palate'.

Due to the diagnosis, some respondents reported spending less time with their baby after the birth (46%, 165/356). In 23% of cases (81/356), the baby had been taken from the room after the birth, even though no additional complications had been observed.

Following the diagnosis, some respondents had received an explanation of what CL/P could mean for them and their child either immediately (12%, 54/466) or within an hour (8%, 39/466) of diagnosis. However, 29% of respondents (133/466) had been left to wait for over an hour, 27% (126/466) had waited for more than one day and 9% (44/466) had waited for more than three working days. In many cases, a Cleft Clinical Nurse Specialist (54%, 252/467) or another CL/P specialist health professional (such as a cleft surgeon; 9%, 42/467) had provided this explanation.

At the time of diagnosis, although 50 of respondents (227/452) felt that they had received the right amount of information, 41% (187/452) felt they had received too

little. 3% (13/452) felt they had received too much and 6% (25/452) answered didn't know/weren't sure. Further, only 40% (183/453) felt that this information had covered all the right topics and had addressed all the queries they had. Nonetheless, 70% of respondents (319/454) were told where to get further information and support. The source of this information and support most often included a Cleft Clinical Nurse Specialist (70%, 317/453), CLAPA's website (30%, 137/453) and/or a CLAPA leaflet (29%, 131/453).

Further, although the majority of parents reported having had a prior feeding plan of exclusive breastfeeding (55%, 252/459), only five% (23/457) were able to do this. Instead, parents reported offering milk from a bottle; using expressed milk (13%, 59/457), a mixture of expressed and formula milk (13%, 60/457) or formula milk (30%, 135/457), as well as nasogastric tube along with breast or bottle feeding (13%, 58/457).

Quantitative Findings – cleft palate only

76% of respondents (358/470) had received a diagnosis of cleft palate within 24 hours of birth, while 15% (69/470) had received a diagnosis between 24 hours and three weeks after the birth. According to 5% of respondents (24/470), the diagnosis had been given more than three weeks after the birth of their child. In the majority of cases (51%, 239 / 466), the diagnosis had been communicated by the midwife or another nurse.

Parents who had received a 'delayed' diagnosis of cleft palate more than 24 hours after birth reported that before a diagnosis was made they experienced problems feeding their baby effectively (83%, 93/112) and had felt personally responsible for these feeding difficulties (72%, 80/112). In 54% of cases (60/112), respondents had

been discharged from the hospital and sent home before the diagnosis was made. 58% (65/112) reported that they had raised concerns but were told by a health professional that nothing was wrong and a further 72% (81/112) reported that the cleft had been missed by multiple health professionals on many occasions.

It is important to note that years of diagnoses ranged considerably in the current sample (from before 1980 to 2016; refer to Figure 1). However, no significant differences were found in overall levels of satisfaction between parents who received a diagnosis prior to, or after the publication of the 2014 Royal College of Paediatrics and Child Health guidelines in 2014 ($t(445) = .507$, $p < .05$).

Qualitative Findings

Qualitative data was also collected in regard to parents' experiences of postnatal diagnosis and care. These findings are summarised alongside exemplar quotes in Table 3.

Discussion

This study aimed to explore parents' experiences of receiving a postnatal diagnosis of CLP, as well as issues pertaining to neonatal care. The findings provide important insight into the key challenges experienced by parents, in addition to practical suggestions for how to improve the diagnostic experience in practice.

Synthesis of Findings

Unsurprisingly, receiving a diagnosis of CL/P brought about strong emotional reactions for parents, which echoes previous findings (Nelson et al., 2012). The quality of diagnostic information as well as the way in which it is delivered is known to be crucial

for long-term parental wellbeing (Maes et al., 1998; Matthew et al., 1998; Chuacharoen et al., 2009; Vanz & Ribeiro, 2011). However, 39% of the participants in this study reported feeling dissatisfied or very dissatisfied with their overall diagnostic experience. Parents perceived a lack of knowledge, sensitivity, and empathy from non-specialist hospital staff, and reported that outdated terminology had been used to refer to their child's diagnosis (such as ''hare lip''). Further, a considerable number of participants felt that they had received too little information, had been given inaccurate information, or had felt overwhelmed by the amount of information provided.

In the case of isolated cleft palate, a 'delayed' diagnosis can not only cause the family undue stress, but can also impact upon the child's nutrition, weight gain, and the parent-child bond, due to undiagnosed feeding difficulties (Habel et al., 2006; Tierney et al., 2015). Feeding difficulties are not uncommon among children with all types of cleft, yet these challenges and associated consequences are amplified in regard to delayed diagnosis of cleft palate. In the present study, respondents who had received a delayed diagnosis described their pre-diagnosis feeding efforts to be physically and emotionally exhausting, and some felt personally responsible for the feeding difficulties. Once the diagnosis had been made, appropriate feeding equipment was not always immediately available. National guidelines indicate that infants must undergo a comprehensive feeding assessment by a cleft Clinical Nurse Specialist prior to the introduction of assisted feeding, emphasising the importance of timely referrals to cleft teams.

In addition, 10% of participants reported that their child's cleft lip had been missed on one or several occasions during the antenatal pregnancy scans. Given that the national standard is for 75% of cleft lips to be detected antenatally; as smaller clefts of the lip can be more difficult to detect on 2D scanning, and complicated by unfavourable

foetal position and increased maternal habitus; this finding is not particularly alarming. Nonetheless, the potential psychological impact on parents of missing the opportunity for antenatal diagnosis of cleft lip must be acknowledged and appropriately handled. Health professionals need to be aware of the potential psychological impact that a diagnosis at birth may have, particularly in relation to the parents' inability to prepare (Nusbaum et al., 2008), and feel confident in delivering information sensitively, offering emotional support, and signposting parents to reliable external resources.

The findings of this study add to the growing body of CL/P literature in which parents describe dissatisfaction with the interactions they experience with non-specialist health professionals (Johansson & Ringsberg, 2004; Knape et al., 2010; McCorkell et al., 2012; Stock & Rumsey, 2015). This finding has been replicated in related studies of disability and chronic illness (Anderson et al., 2007; Smith et al., 2015), rare disorders (von der Lippe, 2017), and other congenital conditions (Wool & Catlin, 2011). While hospital staff cannot be expected to possess expert knowledge on all of the health conditions they treat, the consequences of delivering inaccurate or partial information in a way that is perceived to be insensitive can be considerable and long-lasting, both in relation to the psychological adjustment of the family, and to the physical, cognitive, and social development of the child (Anderson et al., 2007; Habel et al., 2006; Maes et al., 1998; Matthew et al., 1998; Chuacharoen et al., 2009; Vanz & Ribeiro, 2011; Wool & Catlin, 2011). Health professionals need to be aware that parents may be distressed and disoriented and as such require the experience and skill to determine the level of need of each individual and to respond accordingly (Schuth et al., 1994; Lalor et al., 2008). However, due to recruitment and retention issues in maternity units, this would necessitate frequent and regular training in each unit to ensure equal awareness among all staff who may come into contact with babies

affected by orofacial clefts. Targeting such training in relation to Continuing Professional Development, as well as at undergraduate level would help to address this. A foundation level of training on how to deliver diagnoses in a sensitive manner, in a written, online and/or face-to-face format has been written to support health professionals to deliver challenging news (Lalor et al., 2008), and ensure that health professionals are able to provide up-to-date and accurate information confidently, to the level of their degree of specialism. Although this level of training is becoming more prevalent (Royal College of Paediatrics and Child Health, 2014), more could be done to ensure all trainee and qualified professionals are appropriately equipped and supported to deliver optimal care. Further research and Public Involvement activities are needed to guide the development and evaluation of such training and materials.

Many participants reported a high level of satisfaction with specialist CL/P cleft teams, once a diagnosis had been made. This finding replicates previous work (Berggren et al., 2012; Johansson & Ringsberg, 2004; Stock & Rumsey, 2015), and suggests that closer links between local hospitals and specialist services could help to close gaps in the knowledge of congenital conditions such as CL/P, and ensure the timely delivery of reliable information and optimal neonatal care in primary and secondary health care settings.

It is imperative to fully consider the limitations of the current study. First, the survey was advertised through CLAPA's website and social media platforms, thus only reaching parents receiving information and/or services from CLAPA. While CLAPA is the only dedicated national CL/P charity and has a considerable membership base, it cannot be assumed that respondents are fully representative of the population. Further, it is possible that the subgroup of parents who volunteered to take part in this study share a similar set of traits or characteristics which could make them more likely

to volunteer their experiences. Indeed research suggests that those most willing to discuss their experiences might represent extremes; having had particularly good or particularly bad experiences; potentially resulting in a sampling bias (Peel, Parry, Douglas & Lawton, 2006). Additionally, the sample for this study consisted predominantly of White mothers. The lack of a more diverse sample means that some parents' experiences may have been missed. A lack of a representative samples is not uncommon in the health literature and critics note that much of the knowledge base in relation to CL/P is dominated by (often White) mothers' views and experiences (Stock & Rumsey, 2015; Zeytinoglu et al., 2016). Further, the survey required parents to provide retrospective information about their experiences, which means the study is subject to recall bias. Not all respondents answered all the questions on the survey, meaning that some data is missing.

In the current sample, the years of diagnoses range considerably; from before 1980 up until 2016 (see Figure 1). Nonetheless, no significant differences were found in overall levels of satisfaction between parents who received a diagnosis prior to, or after the publication of the 2014 Royal College of Paediatrics and Child Health guidelines in 2014. These findings demonstrate that delayed diagnosis and the neonatal care of infants with a cleft is still a significant concern for at least some parents. However it is imperative to note that guidelines such as these take time to implement (McElroy et al. 2017), and the sample included in this study cannot be deemed to be nationally representative. National initiatives, such as the CRANE database (www.crane-database.org.uk) and the Cleft Collective Cohort Studies (Stock et al., 2016) will be better placed to address these questions in the future.

In spite of these limitations, the present survey was able to capture a large amount of mixed-methods data, which add weight to the importance of timely diagnosis, positive

relationships between families and health professionals, and the delivery of appropriate information and support.

Conclusions

The findings of this study provide important insight into the diagnostic and neonatal care experiences of parents of children born with CL/P. Several service-related issues, including delayed diagnosis of cleft palate, parental dissatisfaction with the delivery of information and support, and the physical and psychological impacts of parental distress were identified. A foundation level of standardised training for primary and secondary care health professionals is therefore recommended.

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