Breaking the news: Parents' experiences of receiving an antenatal diagnosis of cleft lip

Abstract

Objective: In high-income countries, many cases of cleft lip are now identified at the 18-21 week pregnancy scan. The manner in which a diagnosis is communicated is vital for long-term parental wellbeing, yet previous studies have been indicative of parental dissatisfaction. The aims of the present study were to examine the experiences of parents who received an antenatal diagnosis of cleft lip in their unborn child, and to offer pragmatic suggestions for improving the diagnostic experience in practice.

Design: An online, mixed-methods survey was designed and distributed by the Cleft Lip and Palate Association. Data from 574 eligible parents were analysed using descriptive statistics and inductive content analysis.

Results: Although survey responses indicated positive diagnostic experiences overall, respondents perceived a lack of sensitivity among sonographers, long waiting times between referrals, and a lack of appropriate and reliable information. Respondents also reported a number of misconceptions about cleft lip and/or palate and its prognosis, as well as a variety of initial concerns about their own ability to cope with the anticipated challenges.

Conclusion: Findings emphasise the importance of providing accurate and individualised information to prospective parents, in a sensitive manner, so they can adjust to their child's diagnosis and prepare for the birth appropriately. Given that antenatal screening for cleft lip is becoming more fully integrated into routine practice, more training for healthcare professionals, improved access to reliable information in a variety of formats, and stronger links between local hospitals and specialist cleft services may be needed.

Keywords: diagnosis; cleft lip; maternal health; ultrasound; sonography; pregnancy

Introduction

A cleft lip and/or palate (CL/P) is one of the most common congenital conditions in the world, affecting one in every 500-700 live births each year (World Health Organization, 2012). While the initial surgical repair usually takes place during the first year of life, the child will be expected to engage in a long-term multidisciplinary treatment pathway. Research has demonstrated the potential impact of CL/P and its treatment on the child's social, emotional, and cognitive development, as well as on the wellbeing of the parents and the family as a whole (Stock & Feragen, 2016; Nelson et al., 2012). In addition, approximately 30 percent of all cases of cleft lip and palate, and 50 percent of cases of cleft palate alone are considered to be syndromic (Dixon et al., 2011). Common genetic syndromes associated with CL/P, which carry additional medical and psychosocial challenges, include Pierre Robin sequence, Stickler's syndrome, and 22Q11.2 deletion syndrome. In rare cases, a cleft may be indicative of more serious problems, including Down's, Edwards' or Patau's syndromes, which may lead to potentially fatal complications.

In high-income countries, fetal anomaly screening at 18-21 weeks now routinely includes screening for a cleft lip (e.g. Public Health England, 2013). If detected, prospective parents are typically given an appointment for a second scan to confirm and discuss the diagnosis with a maternal-fetal medicine consultant. Following confirmation of a cleft lip, a referral should be made to the local specialist cleft team within 24 hours (e.g. NHS England, 2013). National charities dedicated to helping those born with CL/P and their families, such as the Cleft Lip and Palate Association (CLAPA; United Kingdom) and the American Cleft Palate-Craniofacial Association (United States) also provide specialist information and peer support following a diagnosis.

While an antenatal diagnosis can be distressing (Hsieh et al., 2013; Nusbaum et al., 2008), it also allows prospective parents to adjust to their child's condition and engage with specialist services ahead of the birth (Smith et al., 2015; Berggren et al., 2012; Kuttenberger et al., 2010; Robbins et al., 2010; Rey-Bellet & Hohlfeld, 2004; Aspinall, 2002; Moss, 2001; Davalbhakta & Hall, 2000). To promote optimal long-term parental wellbeing, the manner in which the diagnosis is communicated

is vital (Vanz & Ribeiro, 2011; Chuacharoen et al., 2009; Maes et al., 1998; Matthew et al., 1998). Although several studies in the field of CL/P have acknowledged concerns regarding the way in which antenatal diagnosis is handled (Nelson et al., 2012), relatively few studies have specifically investigated parents' diagnostic experiences and their suggestions for how the service may be improved. This is particularly important given that the validity of ultrasound diagnosis is continuing to evolve (Johnson & Sandy, 2003). The objectives of the present study were to utilise an online, mixed-methods approach to examine the views and experiences of parents who received an antenatal diagnosis of cleft lip in their unborn child, and to offer pragmatic suggestions for improving the diagnostic experience in practice.

Method

Design

An online, mixed-methods survey was designed by CLAPA using the online survey platform, SurveyMonkey. 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 ethical approval was obtained from the Faculty Ethics Committee at the University of the West of England, Bristol, and adhered to at all times. The study was also conducted within the ethical guidelines of the British Psychological Society. Respondents were made aware that their contribution to the survey was voluntary, that their data would be kept confidential, that they would not be personally identified during dissemination, and that they could withdraw their data from the study at any time prior to publication. The survey was advertised on the charity's website, e-

newsletters and social media between October 2016 and January 2017. A total of 1,044 eligible survey responses were collected. Of these, 574 respondents reported receiving an antenatal diagnosis of cleft lip. The present manuscript describes the quantitative and qualitative data provided by these 574 respondents specifically on the topic of antenatal diagnosis. Quantitative data were analysed using descriptive statistics. Since not every respondent answered all of the survey questions, reported percentages were adjusted depending on the number of answers received. Qualitative data were analysed by the second author using inductive content analysis. This type of analysis is deemed appropriate when the aim of a study is to succinctly summarise a large body of qualitative data, and when existing theory or research literature on a given phenomenon is limited (Neuendorf, 2017). First, the data were read and re-read, to establish an overall picture of the data. Common themes were then inductively grouped together in an iterative process (Neuendorf, 2017). A proportion of the data was also coded by the first author to assess reliability. Qualitative codes were then compared, with initial coding reaching an average agreement of 95% (range 93-100%). Any discrepancies were subsequently discussed 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 574 parents who participated in this study. Figure 1 also provides a visual representation of the year of diagnosis.

Quantitative findings

All survey responses included in this study had received a diagnosis of cleft lip during (*n* = 527/574, 92%), or shortly after (*n* = 47/574, 8%) the antenatal anomaly scan. In the majority of cases, the sonographer had delivered the initial diagnosis (*n* = 495/572, 87%). Following the initial anomaly scan, most respondents reported undergoing a second scan to confirm the diagnosis (*n* = 448/568,

79%); however, only 29% had received their second scan within three working days (*n* = 164/568). In 35% of cases (*n* = 199/567), respondents had been informed about other conditions related to CL/P, such as Stickler's syndrome and 22Q11, while 35% of respondents (*n* = 199/567) had also been told about potentially fatal conditions linked to cleft palate, such as Edwards' or Patau's syndrome. Most respondents felt that the diagnosis was presented in either a very sensitive (*n* = 219/572, 38%) or somewhat sensitive (*n* = 224/572, 36%) manner. However, 10% of respondents felt that the diagnosis had been presented insensitively (*n* = 58/572) and a further 12% believed the diagnosis had been delivered very insensitively (*n* = 66/572). Overall, the majority of respondents reported being either very satisfied (*n* = 197/574, 34%) or satisfied (*n* = 178/574, 31%) with their diagnostic experience. Nonetheless, fifteen percent of respondents (*n* = 84/574) remained neutral, 12% (*n* = 67/574) reported being dissatisfied, and 8% (*n* = 48/574) reported being very dissatisfied. No differences in overall satisfaction were observed between respondents who received a diagnosis prior to the implementation of the guidelines in 2010 and those who received a diagnosis after 2010.

Prior to receiving information about the diagnosis, respondents reported having a number of common misconceptions about CL/P. This included the idea that CL/P is purely cosmetic (*n* = 151/554, 27%), that CL/P has a solely hereditary aetiology (*n* = 131/554, 24%), that all children with CL/P will have learning difficulties (*n* = 77/554, 14%), that a diagnosis of CL/P means the child will be disabled (*n* = 63/554, 11%), and that CL/P only affects infants in low-income countries (*n* = 20/554, 4%). Fifty-seven percent of respondents (*n* = 313/553) felt that they had received the right amount of information at the time of diagnosis, while 37% (*n* = 202/553) felt they had not received enough, and 1% (*n* = 8/553) thought the amount of information given was too much. Respondents' greatest concerns following the diagnosis (besides the physical health of their baby) included: other peoples' reactions to their child looking and/or sounding different (*n* = 358/561, 64%), being able to feed the baby (*n* = 336/561, 60%), entrusting the child to the surgical team for repair procedures (*n* = 204/561, 36%), coping with the long-term treatment pathway (*n* = 147/561, 26%), others treating them or their child differently (*n* = 127/561, 23%), concerns about whether they or their partner would be able to bond

with their child (*n* = 116/561, 21%), having to tell friends and family about the diagnosis (*n* = 87/561, 16%), having to explain CL/P to their other children (*n* = 19/561, 3%), and having to tell their partner about the diagnosis (*n* = 4/561, 1%). Although the majority of respondents (*n* = 446/551, 81%) reported that they had been told about where to seek further information and/or support, only 45% (*n* = 246/551) felt that the information they had received had covered the right topics and had addressed all the queries they had.

Qualitative findings

In addition to these quantitative data, 298 qualitative responses about parents' experiences of antenatal diagnosis were extracted. Parents' responses related to the emotional impact of the diagnosis, information provision, experiences with healthcare professionals, waiting times, and experiences with cleft specialists. These findings are summarised in Table 3.

Discussion

Synthesis of Findings

This study provides crucial insight into the antenatal diagnostic experience of cleft lip as perceived by parents. While the findings of this online, mixed-methods survey indicate positive diagnostic experiences overall, respondents also perceived a lack of knowledge and sensitivity among sonographers, long waiting times between referrals, and a lack of availability of appropriate and reliable information. Respondents reported a number of misconceptions about CL/P and its prognosis, as well as a variety of initial concerns about their own ability to cope with the anticipated challenges. Subsequent engagement with specialist CL/P services was reported to be very positive.

Responding to Parents' Emotional Needs

Previous studies have acknowledged the emotional impact of an antenatal diagnosis and identified a perceived lack of empathy from healthcare professionals in response (Lalor et al., 2007; Schuth et al., 1994). Healthcare professionals need to be aware that parents may be distressed and disoriented, and

require experience and skill to determine the level of need of each individual and to respond accordingly (Lalor et al., 2008; Schuth et al., 1994). This is especially relevant given that parents may struggle to absorb information when they feel overwhelmed, as has been demonstrated in the present study. Rather than using medical or outdated terminology, a comprehensible description of the condition, its treatment and its prognosis should be provided (Aspinall, 2002; Schuth et al., 1994).

Information Provision

The quality of the information received at the time of diagnosis can considerably influence parents' long-term wellbeing (Vanz & Ribeiro, 2011), as well as their attitudes toward the diagnosis, further antenatal testing, and even termination of the pregnancy (Stock et al., 2018; Brajenović-Milić et al., 2008). Specialist CL/P materials should be on hand for parents at all stages of the diagnostic pathway. To suit different parents' needs and to enhance accessibility, information should be available in a variety of formats (e.g. leaflets, trusted websites). Information could also be provided cumulatively, to ensure the amount, content and timing of information is optimal for each individual's coping style (Lalor et al., 2008).

Training for Healthcare Professionals

Basic training, possibly in the form of written, online or face-to-face 'Continuing Professional Development' certification, could be beneficial in supporting healthcare professionals to deliver difficult news (Lalor et al., 2007), and to ensure that healthcare professionals are able to provide up-to-date and accurate information confidently, to the level of their degree of specialisation. Future research and public involvement activities are needed to guide the development and evaluation of such training, and work toward this goal is currently underway. In addition, improved links between local hospitals and specialist CL/P services is necessary, so that specialist emotional support and realistic, balanced accounts of what life is like with a child with CL/P can be offered (Aspinall, 2002), and so that any gaps between referrals can be appropriately bridged.

Methodological Considerations

Limitations of the present study must be acknowledged. Specifically, the survey was only shared with parents who are a part of CLAPA's community. While CLAPA's community is considerable, it cannot be assumed that this group, nor the subgroup who responded to the survey, are representative of the CL/P population as a whole. 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 more complex questions in the future, such as pre-post improvements in the diagnostic experience. Further, not all survey respondents answered all of the questions, and therefore some data is missing. Additional questions which may have shed further light on the findings, such as socioeconomic status were not included in the survey, and should be considered for inclusion in future work. A mismatch is also evident between the results of the quantitative analysis and the codes identified in the qualitative data, with the qualitative findings painting a slightly more negative picture overall. It is possible that those who recalled a particularly negative experience were more compelled to provide further details of their experiences. It is also possible that some of the answers given by participants were subject to recall bias. All individuals reshape their life stories in an attempt to make meaning out of their experiences, and it could be that participants emphasised or diluted certain parts of their story. However, this does not make their recalled experiences any less significant. In spite of these limitations, this survey provided a large amount of quantitative and qualitative data on a subject which has to date received relatively little attention in the context of CL/P. The findings will be used to inform future research in this area, and are pertinent to the ways in which antenatal care is delivered in clinical practice.

Conclusions

Taken together, the findings of this and other studies emphasise the importance of providing accurate and individualised information to prospective parents, in a sensitive manner, so that they can adjust to their child's diagnosis and prepare for the birth appropriately. Given that antenatal screening for cleft lip is becoming more fully integrated into routine practice, the results of the current study suggest

that more training for healthcare professionals, improved access to reliable information in a variety of formats, and stronger links between local hospitals and specialist CL/P services is needed.

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