Treatment Experiences in Adults Born with Cleft Lip and/or Palate: A Whole of Life Survey in the United Kingdom

Abstract

Objectives: Adults with cleft lip and/or palate (CL/P) may report dissatisfaction with aesthetic and/or functional outcomes and express desire for further treatment. Although medical intervention can improve quality of life, surgical procedures require complex decision-making and can invoke psychological distress. The aim of this study was to gain a better understanding of ongoing treatment-related support needs by examining self-reported treatment experiences of a sample of adults born with CL/P.

Design: An online, mixed-methods survey was designed by the Cleft Lip and Palate Association in collaboration with (*University*). A total of 207 eligible responses were received. Qualitative data were analysed using inductive content analysis, while quantitative data were analysed using descriptive statistics.

Results: Although participants were satisfied with surgical and dental/orthodontic outcomes overall, many reported ongoing difficulties. Some stated they would seek further treatment while others felt conflicted about risks and benefits. Some had refused treatment, citing treatment fatigue, anxiety, and/or concerns that treatment would be unsuccessful. More than 40 percent of participants were unaware of their entitlement to cleft-related treatment provided by the National Health Service, and many others had experienced difficulties accessing care.

Conclusions: While advances in healthcare may offer opportunities for adults to improve their quality of life, treatment decisions should be weighed with the support of a clinical psychologist where available. Guidance for young adults considering future treatment is also recommended. Finally, training and resources for local practitioners may improve general awareness of CL/P services and increase access to specialist care for adults with ongoing CL/P-related concerns.

Key words: cleft lip and palate; adult; treatment; decision-making; surgery; orthodontics; restorative dentistry

Introduction

For individuals born with a cleft lip and/or palate (CL/P), long-term multidisciplinary treatment is a part of life. Primary repairs during infancy and other early interventions are considered necessary to improve function and optimise aesthetic outcomes. However, as the child grows older, many procedures and treatments become optional and the decision-making process becomes more complex (Wogden et al., 2019). Several studies have demonstrated a high level of patient satisfaction following intervention (Stock & Feragen, 2016), with patients reporting they would undergo the same treatment again knowing the result (Byrne et al., 2014) and that they would recommend the treatment to others (Sharp et al., 2008). Yet, studies have also highlighted that treatment results may not always be seen to outweigh the associated burden or discomfort (Alansari et al., 2014; Stock et al., 2015). Further, patients may feel anxious about or intimidated by treatment (Bos & Prahl, 2011; Hall et al., 2012), find treatment information difficult to understand (Noor & Musa, 2006), and/or feel pressured to comply with treatment recommendations (Hall et al., 2012). Some studies have identified an association between a higher number of surgeries and decreased psychological wellbeing and quality of life (Broder et al., 2012; Wehby et al., 2012). This may reflect cases of greater medical complexity which may necessitate further treatment.

For adults returning to the CL/P service after many years, treatment advances may offer new opportunities for functional and aesthetic improvements. In particular, adults with CL/P often report dissatisfaction with the appearance of the nose, upper lip, facial profile, and teeth (Chuo et al., 2008; Versnel et al., 2010), as well as a range of functional difficulties with the potential to impact on everyday quality of life (Gkantidis et al., 2015; *manuscript in press*). As a result, around 45 percent of adults with CL/P express a desire for further treatment (Marcusson et al., 2002; Sinko et al., 2005; Kappen et al., 2019). However, there is also potential for significant psychological distress and disappointment if adults' expectations of treatment outcomes are not met (Stock et al., 2015; Kappen et al., 2019). Calls have therefore been made for additional investigation into the ongoing treatment needs of adults (see Stock & Feragen, 2016) in order to better understand adults' motivations for treatment, to facilitate psychological wellbeing, and to better support the complexities of the decision-making process. The aim of the current study was therefore to examine the self-reported treatment experiences of a national sample of adults born with CL/P.

Method

Design

Drawing upon previous literature, an online, mixed-methods survey was designed by the Cleft Lip and Palate Association (CLAPA), the UK's largest charity supporting people and their families affected by cleft across the lifespan, in collaboration with (*University*) using the online survey platform, SurveyMonkey. The survey consisted of quantitative questions including multiple choice questions, and five-point rating scales, and open-ended qualitative questions where participants could enter free text. The survey was also available in paper format contained within a 54-page booklet. The survey consisted of 220 questions split across 12 sections. The survey was conducted as part of a larger programme of work aimed at improving the support available to adults born with CL/P in the UK (the CLAPA Adults Services Programme). Given the volume of data collected from the survey, findings are reported across a series of papers, of which the current paper is one. This paper reports on the data from 32 of the 56 questions in section 2 titled "Your Health" of the survey, supported by demographic data from sections 1, 6, 8 and 11. The remaining data is reported in other papers (see table 1).

Materials

The survey was designed using current literature (e.g. Stock & Feragen, 2016) and the expertise of the research team. The question types included questions rated on a 5-point Likert scale, such as "How much do you agree with the following statements? (1 = strongly disagree; 5 = strongly agree)", as well as open-ended questions, such as "In your own words, please summarise why you feel those relationships ended". Weighted averages are also used when reporting some responses. Weighted averages give an indication of how strongly a respondent feels about a particular statement relative to other statements by assigning each item a 'weight' based on the frequency with which it is selected and therefore reflects the relative importance of each item. The advantage of this approach is that rather than results being biased toward the 'middle' if there is wide variation, a weighted average depicts where the majority of the observations fall. The draft survey was piloted with the CLAPA Adult Voices Council (AVC), a group of eight adults born with CL/P. The AVC examined all proposed questions to ensure readability and that the results could be interpreted in a way which would answer the question that the researchers sought to determine. Questions which were unclear were either removed or amended with input from the AVC. The final version was later piloted with six self-selecting volunteers (all adults living in the UK who were born with CL/P).

Procedure

Institutional ethical approval was obtained from the Faculty Ethics Committee at (*University*). The survey was advertised via direct e-newsletters specifically promoting the survey to CLAPA's members; announcements made during interviews in national and local media; posters and leaflets; and CLAPA's social media between July and October 2018. Prior to survey completion, potential participants 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. Participants were asked to indicate their consent for their data to be used for research purposes. According to participant preference, surveys were completed either online (via the platform SurveyMonkey) or in paper format contained within a 54-page booklet.

Analysis

Qualitative data were analysed independently by the first and fourth authors using inductive content analysis (Elo & Kingas, 2008). 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. Initial codes were generated for each comment and notes were kept throughout (Hsieh & Shannon, 2005). Next, codes were inductively grouped together into broad categories (e.g. 'Concerns with Surgical Outcomes'; Dey, 1993). Following this, sub-categories were created (e.g. 'Breathing/Sinus Issues'; Dey, 1993) and frequency counts were calculated. Any discrepancies in coding were discussed between the first and fourth authors until full agreement was reached. The qualitative data presented in the current paper relates to the main category of 'Treatment Experiences'.

Quantitative data were analysed by the first and fourth authors. Descriptive statistics were used to analyse data collected from survey questions. Since most questions were optional, not every participant answered all survey questions. Reported percentages were therefore adjusted depending on the number of responses received. In many cases, participants were able to select more than one answer, and some added totals therefore exceed 100%. Figures were rounded to one decimal place.

Results

Participants

A total of 224 responses were received. Seventeen participants were excluded as they had been born outside of the UK (7.6%). Of the remaining 207 eligible responses, 94.7% were completed online. A further 5.3% returned paper versions via post. These data were entered into Survey Monkey by a student intern and all surveys were checked by the first author. The most commonly reported recruitment method was a CLAPA e-newsletter campaign (44.4%). Other successful recruitment methods included social media (32.8%), direct contact with CLAPA staff (8.9%), word of mouth (3.9%), posters and leaflets (3.3%), and the CLAPA website (2.8%).

A total of 207 eligible participants contributed data to the questions which are reported in the present paper. Participant demographics are provided in Table 2, alongside National census data where available (Cleft Registry and Audit Network, 2018; Office for National Statistics, 2018).

Surgical Experiences

Participants reported having undergone the following surgical procedures during adulthood: rhinoplasty (*n* = 107; 53.2%), orthognathic surgery (*n* = 47; 23.4%), pharyngoplasty or pharyngeal flap (*n* = 38; 18.9%), fistulae repair (*n* = 14; 7%), and/or secondary lip revision (*n* = 10; 5.0%).

Participants were asked to respond to a series of statements about their surgical experiences on a scale of 1 (Strongly Disagree) to 5 (Strongly Agree). Weighted averages were calculated for each statement and are presented in order from high to low: *"Overall, I am happy with the surgical outcomes from my cleft treatment"* (4.0); *"Overall, I am pleased with the range of surgeries that I was offered"* (3.9); *"Overall, I am pleased with the surgical care and follow-up that I received"* (3.9); and *"Overall, I feel that my surgical team listened to me, understood my concerns, and acted upon my concerns"* (3.8).

Experiences of General Dental Treatment, Orthodontics, and Restorative Dentistry

A total of 147 participants (77.0%) had visited a General Dental Practitioner (GDP) within the last 12 months. Twenty-three participants (12.0%) had last visited a GDP between one and two years ago. A minority hadn't visited a GDP for two to five years (*n* = 9; 4.7%), five to 10 years (*n* = 6;

3.1%), or more than 10 years (*n* = 1; 0.5%). Five participants couldn’t remember the last time they had visited a GDP (2.6%). A total of 141 participants reported concerns about visiting the dentist. These concerns included: cost (*n* = 79; 56.4%), fear/anxiety (*n* = 70; 50.0%), pain/sensitivity (*n* = 54; 38.6%), a lack of understanding of CL/P among GDPs (*n* = 48; 34.3%), and having had a prior bad experience (*n* = 30; 21.4%). Nineteen participants (9.8%) reported having ongoing difficulties accessing general dental services. Participants reported a weighted average of 3.5 when asked to respond to the following statement on a scale of 1 (Strongly Disagree) to 5 (Strongly Agree): *“Overall, I feel that my general dental practitioner listened to me, understood my concerns, and acted upon my concerns”*.

Participants reported having had a range of general dental/orthodontic/restorative dentistry procedures in adulthood. These most commonly included: braces (*n* = 160; 82.5%), tooth extractions (non-wisdom teeth, *n* = 156; 80.4% and wisdom teeth, *n* = 56; 28.9%), and fillings (*n* = 143; 73.7%). Other procedures included: crowns/caps (*n* = 79; 40.7%), having an appliance such as an obturator appliance/quad helix (*n* = 60; 30.9%), bridge/implant (*n* = 58; 29.9%), bonding (*n* = 49; 25.3%), and dentures (*n* = 48; 24.7%).

Participants were asked to respond to a series of statements about their general dental/orthodontic/restorative dentistry experiences on a scale of 1 (Strongly Disagree) to 5 (Strongly Agree). Weighted averages were calculated for each statement and are presented in order from high to low: *“Overall, I am happy with the dental and orthodontic outcomes from my cleft treatment”* (3.7); *“Overall, I am pleased with the range of dental procedures that I was offered”* (3.7); *“Overall, I am pleased with the dental/orthodontic care and follow-up that I received”* (3.6); and *“Overall, I feel that my dental/orthodontic team listened to me, understood my concerns, and acted upon my concerns”* (3.6).

Desire for Further Treatment

Most participants were not scheduled for any further operations at the time of survey completion (*n* = 163; 89.1%). However, a total of 108 (53.7%) participants reported ongoing concerns with their surgical outcomes. These are presented in Table 3. A minority of participants were waiting to undergo a range of procedures, including: rhinoplasty (*n* = 13; 7.1%), secondary lip revision (*n* = 11; 6.0%), fistulae repair (*n* = 6; 3.3%), pharyngoplasty or pharyngeal flap (*n* = 3; 1.6%), and/or orthognathic surgery (*n* = 2; 1.1%). Participants were asked to respond to two statements regarding potential future surgery on a scale of 1 (Strongly Disagree) to 5 (Strongly Agree). Weighted averages were calculated for each statement and are presented in order from high to low: *"I would consider having further surgery if it would improve function"* (3.6); and *"I would consider having further surgery if it would improve my appearance"* (3.4).

A total of 87 participants (45.3%) reported ongoing concerns with their general dental/orthodontic/restorative dentistry outcomes. These are presented in Table 3. Participants were also asked about future general dental/orthodontic/restorative dentistry treatment, as follows: *"I would consider having further dental/orthodontic treatment if it would improve function"* (3.7), and *"I would consider having further dental/orthodontic treatment if it would improve my appearance"* (3.7).

However, 63 participants (35.0%) reported having been offered surgery which they later chose not to undergo. A further 16 participants (8.3%) reported having been offered a general dental/orthodontic/restorative dentistry procedure which they later refused. Reported reasons for these decisions include, for example, being content with current appearance and/or function, the costs outweighing the benefits and having had enough of surgery. A frequency count of all the given reasons are shown in Table 4.

Awareness of Entitlement to NHS Services

Seventy-seven participants (41.4%) were unaware that they were eligible to receive CL/P-specific treatment as part of the NHS. Further, 31 participants (16.7%) had paid privately for CL/P-related treatment and/or therapy as an adult. This most frequently related to general dental/orthodontic/restorative dentistry work (*n* = 23). Seventy-five participants (41.0%) reported feeling concerned that their eligibility to access future services may change in the future.

Discussion

The aim of this study was to examine the self-reported treatment experiences of a national sample of adults born with CL/P. The results offer insight into the treatment status of this under-researched population. The findings are discussed below in more detail, alongside suggestions for further research, and clinical and community practice.

Satisfaction with Treatment Outcomes and Ongoing Concerns

A considerable proportion of participants had undergone surgery in adulthood. In line with previous research (Sinko et al., 2005; Chuo et al., 2008), rhinoplasty was the most commonly sought operation, with just over half of participants reporting they had undergone this type of surgery. Participants also reported undergoing a range of general dental/orthodontic/restorative dentistry procedures. The most commonly cited procedures included braces, extraction of non-wisdom teeth, and fillings. On the whole, participants were reasonably satisfied with the outcomes of their surgical and general dental/orthodontic/restorative dentistry treatment. However, many had ongoing concerns (Table 3). The most frequently reported concerns included facial asymmetry, prior dental work failing, and lip scarring. These findings are in line with previous research stating that while the majority of adults express overall satisfaction with treatment outcomes, lower levels of satisfaction are often found in relation to particular facial features, including the nose, upper lip, facial profile, and teeth (Chuo et al., 2008; Versnel et al., 2010).

In addition to aesthetic concerns, participants reported a number of ongoing functional issues, with the potential to impact long-term health and everyday quality of life. These most commonly included breathing/sinus issues, difficulties eating and drinking, and/or facial pain/numbness. The presence of these difficulties has been previously reported in the literature (Chuo et al., 2008; Gkantidis et al., 2015; *manuscript in press*), yet further research to identify the specific impact of these concerns or ways of improving patients' quality of life is scarce. Some of these difficulties are challenging to address through surgery (e.g. residual fistulae) and thus may require other forms of treatment. For example, some adults will have undergone a general dental/orthodontic/restorative dentistry procedure (e.g. a palatal obturator or speech bulb) for the purposes of improving speech (*manuscript in press*). Additional research to identify ways of supporting adults with these issues is therefore recommended.

Medical Decision-Making in Adulthood

Previous research has suggested that around 45 percent of adults with CL/P express a desire for further treatment (Marcusson et al., 2002; Sinko et al., 2005; Kappen et al., 2019). Unfortunately, research has also identified a greater prevalence of appearance dissatisfaction (Marcusson et al., 2002; Chuo et al., 2008), a higher degree of perceived stigmatisation (Bemmels et al., 2013), and elevated levels of anxiety and depression (Ramstad et al., 1995; Marcusson et al., 2002; Sinko et al., 2005) in those adults seeking surgical intervention. Prior research has identified improvements in psychological wellbeing following surgical intervention (e.g. Hens et al., 2011; Byrne et al., 2014), suggesting that further treatment may be of benefit to some, particularly given that outcomes have significantly improved since today's adults were originally treated (Al-Ghatam et al., 2015; Smallridge et al., 2015). However, qualitative literature has also highlighted the potential for significant distress and disappointment if adults' expectations of surgical outcomes are not met (Stock et al., 2015; Kappen et al., 2019). In the current study, some participants stated they would opt for further aesthetic/functional treatment and/or were already on the surgical waiting list, while others felt more conflicted about the relative risks and benefits. In line with previous research (Hall et al., 2012; Alansari et al., 2014; Krikken et al., 2015; Stock et al., 2015), a proportion of participants had refused further treatment in adulthood, citing concerns that treatment would be unsuccessful, treatment fatigue, and/or treatment anxiety stemming from unpleasant childhood experiences. Given that most participants would have received the majority of their treatment prior to the introduction of clinical psychologists to UK cleft teams, it is doubtful that they would have had opportunity to discuss and address any psychological concerns. To avoid unnecessary treatment-related distress in adulthood, these findings emphasise the need for treatment decisions to be carefully weighed; balancing what is surgically possible with the patients' treatment history, motivations, and expectations. The potential treatment burden and the patient's individual circumstances should be taken into consideration, with the involvement of a clinical psychologist and the use of psychological screening tools where available (Aspinall, 2010; Cadogan & Bennum, 2011; Wogden et al., 2019). Finally, several participants had refused further treatment, citing positive self-perceptions as a reason for no longer needing corrective treatment; a finding also identified by previous research (Stock et al., 2015). CL/P teams should therefore be careful not to offer an array of surgical options to adults returning to the service unless this is desired by the patient (Aspinall, 2010), and ensure that the option of not pursuing further treatment is offered alongside other treatment options. In general, providers should be mindful of how education about treatment options are described with use of neutral language so patients are aware of their treatment options with decision making reflecting each patient's individual concerns and goals.

Access to Local and Specialist Treatment

More than 40 percent of participants in the current study were unaware of their entitlement to cleft-related treatment on the NHS. Consequently, some had paid for private treatment. Previous qualitative research has also highlighted these issues, stating the need for access to information, treatment, and support in adulthood, while acknowledging the difficulties for the NHS and its partners in reaching a population who have previously been 'discharged' from the CL/P service (Stock et al., 2015; Stock et al., 2018). This is a particular challenge for those adults who were 'discharged' prior to the centralisation of cleft care and who may therefore be entirely unaware of the existence of specialist CL/P teams. Additionally, they may have been told previously that no further treatment was available for them, even though this may no longer be the case. As a result, they may be less likely to proactively seek care to address any ongoing concerns. Crucially, information should be made available to all young adults who are about to end routine treatment to ensure they are aware of the services available to them and how to access them. Yet, even for those participants who were aware of their entitlement to NHS care, accessing care via a referral from a local health professional had proved challenging for some. In some areas of the UK, a self-referral made directly to the cleft team is possible, yet this is not uniform process across the UK. Previous research has identified a potential lack of knowledge among local health professionals (including GPs and GDPs) regarding CL/P and the corresponding routine treatment pathway (Stock et al., 2015; Stock et al., 2018). Researchers have subsequently called for improved training and resources for local practitioners, closer communication between local practitioners and specialist CL/P teams, and a move by the tertiary sector to inform and empower patients to advocate their eligibility to NHS treatment (Stock et al., 2018).

Just over 40 percent of participants expressed concerns over their eligibility to NHS hospital services changing in the future. Such concerns may reflect the recent decision to centralise CLP services in Scotland, wider cuts to NHS services, and an ever-changing political environment within the UK and internationally. Indeed, even in currently commissioned services, access to restorative dentistry is variable across the UK. Given that such concerns exist even within the NHS framework which does ensure access to care at any age, it is probable that in other countries without such health care coverage, that concerns over eligibility for treatment would be even more profound. Further research with participants living in areas which are reliant on private insurance, for example, would offer a valuable comparison.

Methodological Considerations

Limitations of the present study must be acknowledged. First, the survey was predominantly shared with adults who are existing members of CLAPA. While CLAPA's community is considerable, it cannot be assumed that this group, nor the self-selecting subgroup who responded to the survey, are representative of the UK population. Individuals with cleft palate only were particularly underrepresented in the current sample. How to better represent adults who are less engaged with CL/P services and/or those who are unaware of the services available to them remains a significant challenge. Further, not all participants answered all the survey questions, and therefore some data are missing. Second, survey participants predominantly identified as White and living in England. However, with the exception of the lower participation rate of men, a challenge well acknowledged in studies such as this (Johal et al., 2012), these figures are not considerably different from UK census data (Office for National Statistics, 2018). Nonetheless, several previous CL/P studies have been indicative of poorer outcomes among minority groups (see Stock & Feragen, 2016), and further efforts are needed to ensure that support services are applicable and accessible to the population as a whole. Exploration of the psychological wellbeing and treatment needs of adults who are currently living in the UK but received the majority of their care elsewhere could also be an important consideration for future studies. Multicentre, interdisciplinary, and international collaboration is therefore strongly encouraged to gain a more representative picture of the population and to move toward a better understanding of holistic outcomes in CL/P.

Despite some limitations, this comprehensive survey provides a large amount of quantitative and qualitative data on a group 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 psychological support for adults with CL/P is delivered in clinical practice and in the community.

Conclusions

Adults in the UK with CL/P may experience ongoing issues related to surgical and general dental/orthodontic/restorative dentistry outcomes. Yet, adults may be unaware of the NHS hospital services that are available to them and/or may have difficulties accessing specialist treatment. To address these gaps, information for young adults who are about to complete routine treatment is recommended, as is improved training and resources for local practitioners. The integration of routine patient reported outcome measures from an early age is highly recommended to achieve optimal outcomes in the long term. Comprehensive psychological screening for adults returning to the service later in life is also strongly advised.

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