**Physical Health in Adults Born with Cleft Lip and/or Palate: A Whole of Life Survey in the United Kingdom**

**Abstract**

*Objectives:* Previous research in the field of cleft lip and/or palate (CL/P) has indicated a high prevalence of common health concerns, functional difficulties, and additional conditions known to affect long-term physical health, cognitive development, and psychological wellbeing. The aim of the present study was to examine the self-reported physical health of a national sample of adults born with CL/P.

*Design:* Drawing upon previous literature, 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 between July and October 2018. Qualitative data were analysed using inductive content analysis, while quantitative data were analysed using descriptive statistics.

*Results:* A considerable proportion of participants reported one or more comorbid conditions, including common health concerns, difficulties related to language and/or learning, unresolved speech and/or hearing issues, problems with vision, breathing difficulties, and concerns related to eating and drinking. Many participants were unaware of their entitlement to specialist treatment, and some had experienced difficulties accessing specialist services through their General Practitioner (GP).

*Conclusions:* Individuals with CL/P may be at risk of physical health issues persisting into adulthood. The monitoring of physical symptoms from an early age is recommended, as is a routine physical assessment for adults returning to the CL/P service later in life. Education for both GPs and adults is warranted. Future studies could investigate the wider prevalence of comorbid conditions in CL/P, in order to better understand the longer-term health burden.

*Key words:* cleft lip and palate; adult; physical health; speech; hearing; breathing; self-report

**Introduction**

A long-term condition (LTC) is defined as a health condition that is unlikely to be cured but is controllable through medication and/or other treatment or therapies (Department of Health, 2012). People with LTCs account for 50 percent of all General Practitioner (GP) appointments, 64 percent of outpatient appointments, and 70 percent of inpatient bed days, meaning that 30 percent of the population account for 70 percent of all National Health Service (NHS) spending in the United Kingdom (UK; Department of Health, 2012). These conditions are not only a substantial health issue but can also have a significant impact on an individual’s ability to work and lead a full life (Megari, 2013). As such, the early identification and appropriate management of LTCs is crucial.

Although a cleft lip and/or palate (CL/P) may not be considered a long-term condition in the same sense as diabetes, cancer, or heart disease, it is associated with many ongoing health concerns and comorbidities. For example, adults with CL/P have been found to report difficulties with several functional aspects of the condition. These include concerns with speech and feeding (Oosterkamp et al., 2007; Chuo et al., 2008; Havstam et al., 2011; Munz et al., 2011; Gkantidis et al., 2015; Stock et al., 2015). There are few publications on hearing difficulties in the adult cleft population, but studies suggest that more than 13% of adolescent cleft patients have hearing problems (Narayaran et al., 2013). This is significantly higher than self-reported hearing loss in adolescents in the general population, of 1.0 – 4.0% (, but is lower than up to 90% reported in cleft children under six years (….A high prevalence of developmental difficulties, attention and/or hyperactivity disorders, language impairments, and general learning difficulties with the potential to influence long-term health outcomes have also been identified in the CL/P population (Milerad et al., 1997; Swanenburg et al., 2003; Hyman et al., 2005), in addition to a number of associated syndromes (Feragen et al., 2014; Berg et al., 2016). As such, several studies have concluded that individuals with CL/P are in poorer overall health when compared to the general population (Damiano et al., 2007; Foo et al., 2012; Queiroz Herkrath et al., 2015). Further, large-scale population studies have indicated that individuals with CL/P are at greater risk of psychiatric disease (Christensen and Mortensen, 2002), cerebral palsy (Berg et al., 2016), several types of cancer (Bille et al., 2005), and overall mortality attributable to all major causes of death (Christensen et al., 2004). Taken together, CL/P imposes a potentially considerable economic burden on society (Berk and Marazita, 2002), with an increased number of hospital stays, healthcare use, and related expenditures compared to unaffected individuals (Cassell et al., 2008; Boulet et al., 2009). Understanding the ongoing physical health needs of adults with CL/P is therefore critical for influencing healthcare practices and policies, with the long-term aim of improving health outcomes and reducing the burden at both an individual and societal level (Wehby and Cassell, 2010). In the UK, adults who were born with a cleft are eligible to be seen by the National Health Service (NHS) multidisciplinary cleft team at any age for assessment and/or treatment. Cleft services in adulthood can be accessed via a referral from the patient’s General Practitioner (GP) or General Dentist Practitioner (GDP). In Scotland and Wales, patients are also able to self-refer to the cleft team. The NHS is a state-run healthcare system funded through taxation that is free to patients at the point of access.

With the exception of some population-based studies, individuals with comorbidities are often excluded from research samples. In utilising these exclusion criteria, knowledge of this vulnerable subgroup of patients is lacking (Feragen et al., 2014). Further, the research available tends to focus on children rather than adults, resulting in a loss of understanding of the longer-term outcomes of CL/P. The aim of the current study was therefore to explore the self-reported physical health of a national sample of adults born with CL/P via the administration of a comprehensive survey.

**Method**

***Design***

Drawing upon previous literature, an online, mixed-methods survey was designed by the Cleft Lip and Palate Association (CLAPA) in collaboration with (*University*). The survey is considered to be mixed-methods as it concurrently collected both qualitative and quantitative data. The survey consisted of 220 questions split across 12 sections. An overview of survey content are provided in Table 1. 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).

***Materials***

The survey was designed using the online survey platform, SurveyMonkey. The survey was available online, and also available in paper format contained within a 54-page booklet. Participants self-selected whether they completed the online or paper version. The survey was piloted with the CLAPA Adult Voices Council (AVC). The AVC were a group of eight adults born with a cleft, living in the UK. The AVC examined every proposed question to ensure that it made sense and was interpreted in a way which would answer the question that the researchers sought to determine. Questions which the AVC were unclear on were either removed, or amended with input from the AVC. Following this, the final version was piloted with six self-selecting volunteers (all adults living in the UK who were born with a cleft) in CLAPA’s offices in May 2018.

***Procedure***

Institutional ethical approval was obtained from the Faculty Ethics Committee at (*University*). The survey was advertised via direct e-newsletters to CLAPA’s members; 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. A variety of question types were used. These included multiple choice, for example, “When you are well (i.e. don’t have a cold or other respiratory condition), how do you usually breathe? A. Through my nose (and mouth), B. Only through my mouth, C. Through a stoma/tracheostomy.”, weighted averages, for example, “How much do you agree with the following statement? I am happy with my speech [5 point scale from strongly disagree to strongly agree]”, as well as open ended questions, for example, “Is there anything else that you would like to tell us about your hearing?” Where a medical term was used in a question, a lay summary was also included in parentheses – e.g. “Orthognathic Surgery (Jaw Surgery)”. Where possible, data were compared to national statistics.

***Analysis***

Qualitative data were analysed independently by the first and third authors 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). Qualitative codes were subsequently discussed until full agreement was reached. Finally, frequency counts were calculated.

Quantitative data were analysed by the first and third authors. Descriptive statistics were used to analyse data collected from survey questions. Since most questions were optional, not every participant chose to answer 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. 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.

***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 were entered into SurveyMonkey by a student intern and all surveys were checked by the first author. The most commonly reported recruitment method was a CLAPA email campaign (44.4%). Other successful recruitment methods included social media (32.8%), direct contact with CLAPA staff (8.9%), word of mouth (3.9%), a leaflet or poster (3.3%), or the CLAPA website (2.8%).

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

**Results**

***Comorbid Conditions***

To gain an understanding of where CL/P featured relative to participants’ overall health, participants were asked if they had received a diagnosis of the UK’s most common health conditions. Results are presented in Table 3 alongside general population and published CL/P comparison samples where available. One hundred and fifty-eight (78.6%) respondents did not report a diagnosis of a common health condition, while thirty-two (15.9%) reported a diagnosis of asthma, seven (3.5%) reported a diagnosis of heart disease, six (3.0%) reported a diagnosis of diabetes and four (2.0%) reported receiving a diagnosis of cancer. One hundred and thirty-five participants (67.2%) reported having difficulties with their vision which required glasses or contact lenses to correct.

Similarly, participants were asked if they had received a diagnosis of a condition related to language and/or learning. One hundred and seventy-seven participants (89.4%) did not report a diagnosis, twelve (6.1%) reported a diagnosis of dyslexia, ten (5.1%) reported a diagnosis of language delay, and three reported a diagnosis of dyspraxia (1.5%). Two participants (1.0%) reported a diagnosis of autism spectrum disorder, and one participant reported a learning disability (0.5%). The majority of participants did not report the presence of a syndrome associated with their CL/P (see Table 2). Twenty-two (11.0%) did report having a syndrome, while 31 (15.4%) reported that they didn’t know.

***Breathing***

Participants were asked how they ordinarily breathe when they are well. A summary of results is shown in Table 4. The majority of participants (*n* = 148; 74.8%) indicated they were able to breathe through both their nose and mouth, while fifty participants (25.3%) indicated they were only able to breathe through their mouth. Ninety-six participants (47.8%) stated they found it difficult to breathe at least some of the time as a result of their CL/P. Most participants (*n* = 138; 70.1%) had never undergone a breathing assessment.

A total of 105 (53.6%) participants reported that they snore. Of these, fifteen (7.7%) reported needing to sleep in a different room to others because of their snoring. Only 20 participants (10.2%) had ever seen a doctor regarding their snoring.

Participants were also asked if they woke up three days a week or more with symptoms which could indicate sub-optimal breathing. A large proportion of participants (*n* = 117; 60.9%) reported that they awoke with a dry mouth. Several participants also reported headaches (*n* = 33; 17.2%), light-headedness (*n* = 31; 16.2%), and/or confusion (*n* = 8; 4.2%). Ninety participants (44.8%) reported feeling excessively tired on a regular basis.

***Speech***

Participants were asked if they experienced any ongoing issues with their speech. A summary of results is shown in Table 4. Sixty-four (34.8%) participants reported having no issues, while 55 (29.9%) participants reported hyponasality, 48 (26.1%) reported hypernasality, 38 (20.7%) reported nasal turbulence, and 20 (10.9%) reported having a voice disorder (dysphonia).

Participants were asked to rate how intelligible they believed their speech to be on a five-point scale, where a score of one indicated “*people rarely understand what I am saying”* and a score of five indicated “*people always/almost always understand what I am saying”*. The weighted average for this question was 4.42. Participants were also asked to rate their speech acceptability on a five-point scale where a score of one indicated “*my speech sounds very different and others have commented on this”* and a score of five indicated “*my speech sounds fairly typical for someone who speaks my language and dialect”*. The weighted average for this question was 3.86. When choosing from a list of all English consonant sounds presented orthographically, participants reported that the sound they experienced most difficulties with was /s/, with 47 (27.0%) participants reporting this sound as problematic.

A total of 132 participants (70.6%) reported receiving speech and language therapy (SLT) at some point in their lives. Those who had received SLT were asked to indicate the extent to which they agreed with two statements using a five-point scale (1= strongly disagree; 5 = strongly agree). The weighted averages were as follows: “*Overall, I am happy with the outcomes of the Speech and Language Therapy I received”* (3.70); and “*Overall, I feel that my speech and language therapist listened to me, understood my concerns, and acted upon my concerns”* (3.66). Furthermore, all participants were asked two additional questions, as follows: “*I am happy with my speech”* (3.64); and “*I would consider having [further] speech and language therapy if it would improve my speech”* (3.41).

Some participants provided additional comments regarding their speech. While some stated that they had never received SLT (*n* = 10), others reported having received ineffective SLT (*n* = 5). Three participants had accessed SLT on the NHS in adulthood, while two others had sought private SLT. Some participants felt their speech had improved due to recent surgery (*n* = 4), while others had resolved their speech difficulties through the use of a prosthesis (*n* = 2). Two participants had been told they were ineligible for surgery as an adult but were yet to find an alternative solution.

***Hearing***

A summary of results is displayed in Table 4. A total of 83 (44.6%) participants reported ongoing difficulties with their hearing. Of these, 40 (48.2%) believed that these hearing difficulties were related to CL/P, while 38 (45.8%) reported that they did not know. Sixty-four participants (34.6%) indicated they were experiencing tinnitus. Other reported hearing problems included: mild to severe hearing loss (*n* = 15); middle ear conditions (*n* = 5); a need for hearing aids (*n* = 4); outer ear conditions(*n* = 3); sinus pain (*n* = 2); inner ear conditions (*n* = 1); and Temporomandibular Joint Dysfunction (*n* = 1). Participants also reported that their hearing worsened when ill (*n* = 2) or in the presence of background noise (*n* = 2). Four participants stated they were concerned about their hearing worsening in the future. Of those experiencing hearing difficulties, 31 (40.3%) were under the care of an Audiologist and/or Ear, Nose & Throat specialist (ENT). Some participants reported difficulties accessing an appropriate level of care (*n* = 14).

Participants were asked to review the audiology/ENT care they had received to date. These four questions were presented on a five-point scale from “strongly disagree” (1) to “strongly agree” (5). Weighted averages were calculated and are listed in order from high to low: *Overall I am happy with the outcomes of the audiology treatment that I received* (3.60); *I am happy with my hearing* (3.58); *Overall I feel that my Audiologist/ENT specialist listened to me, understood my concerns, and acted upon my concerns* (3.53); and *I would consider having [further] treatment if it would improve my hearing* (3.43).

***Feeding***

A summary of findings is shown in Table 4. When asked about their experiences with feeding, fourteen participants (8.4%) reported they were unable to eat foods orally and therefore did not complete the remainder of this section. The remaining 153 participants were asked to indicate the extent to which they agreed to a series of statements using a five-point scale from “strongly disagree” (1) to “strongly agree” (5). Weighted averages were calculated and are listed in order from high to low: *I enjoy eating and drinking* (4.37); *I feel that I eat my food in a normal way* (4.12); *I enjoy eating out in restaurants* (4.11); *I am comfortable to eat in front of other people* (3.97); *I am a slow eater* (2.83); and *I find it difficult to eat and/or drink* (1.92).

Nonetheless, 82 participants (49.1%) indicated they often experienced unpleasant symptoms when feeding. These included nasal regurgitation (*n* = 48; 58.6%), reflux (*n* = 29; 35.4%), coughing/choking (*n* = 19; 23.2%), shortness of breath (*n* = 15; 18.3%), general discomfort (*n* = 14; 17.1%), nausea (*n* = 7; 8.5%) and pain (*n* = 4, 4.9%). Forty-one participants (27.2%) also reported avoiding certain foods because of CL/P. These commonly included hard foods (e.g. crusty bread), spicy foods, and small foods that may end up in a fistula (e.g. rice, nuts, seeds, popcorn etc.). Seven respondents (4.6%) reported being unable to drink through a straw, with a further 20 (13.3%) reporting that they can drink through a straw, but with difficulty.

***Engagement with General Practitioners***

A summary of findings is shown in Table 4. The majority of participants had visited a GP within the last twelve months (*n* = 154; 77.0%) and almost all participants (*n* = 198; 99.0%) had visited a GP within the last five years. Not all participants had tried to access CL/P-related services via their GP, but 44 participants (22.0%) indicated that they had experienced difficulties with this. These difficulties included: a lack of GP knowledge about CL/P and its treatment (*n* = 8), the patient themselves being unaware of their entitlement to specialist care (*n* = 8), the GP being dismissive of the patient’s concerns (*n* = 5), struggling to get a tertiary referral processed by primary care (*n* = 12), being referred by the GP to the wrong hospital or department (*n* = 6), the hospital failing to refer the patient on to the specialist CL/P team (*n* = 4), and the patient being referred to costly private treatment (*n* = 1). This had resulted in long waiting times (up to 3 years; *n* = 3) and, in one case, the patient reported receiving treatment that later had to be corrected by the CL/P team. Some participants had learned about their entitlement to NHS via specialist CL/P teams from friends and family (*n* = 2), or via a charity or other route (*n* = 2). Two participants had been successfully referred to the CL/P team by their GP, while two others had successfully handled a self-referral.

**Discussion**

The aim of this study was to explore the self-reported physical health of a national sample of adults born with CL/P in the United Kingdom. To the authors’ knowledge, this is the largest and most comprehensive survey of adjustment in adults to date. The findings provide insight into the ongoing physical health concerns of individuals with CL/P in adulthood and are discussed below in more detail alongside suggestions for further research, and clinical and community practice.

***Frequency of Comorbid Conditions***

In the current study, a considerable minority of participants reported a diagnosis of one or more comorbid conditions (Table 3). These included common health problems, as well as other difficulties known to have a high prevalence in CL/P samples. In particular, the reported rate of asthma was high in adults with CL/P compared to the general population. It is possible that this could represent a tendency to misdiagnose breathing problems, which were also found to be prevalent in the current sample. Although mentioned in previous literature, breathing problems in adulthood have not been well explored, and the findings of the current study suggest this is an area suitable for further research. The reported rate of cancer was found to be at the higher end of the expected figure for the general population but was still within the normal range when taking age into account. While Christensen and Mortensen (2002) identified a high rate of certain cancers in the CL/P Danish population, they were unable to replicate these findings in a follow-up study (Christensen et al., 2004). Population-based studies are challenging to undertake but provide important epidemiological information. To the authors’ knowledge, UK-wide studies on the prevalence of comorbid conditions in CL/P do not currently exist, yet could contribute considerable knowledge about the longer-term health burden of the condition. Vision and hearing problems in the current CL/P sample were both elevated in comparison to the general population, and hearing problems remained high when compared to a Norwegian CL/P cohort (Feragen et al., 2017). This could be due to the relatively older age of the current sample, since vision and hearing problems worsen with age. Nonetheless, both vision and hearing loss remain understudied in the adult CL/P population, and the field could benefit from further research to better understand the long-term impact of these comorbidities and potential interventions to prevent and improve them.

Further, the present study identified a lower-than-expected rate of additional conditions known to affect language and learning, including dyslexia, language impairment, autism spectrum disorder, and learning difficulties when compared to the general UK population. This could be due in part to the format of the study, which involved completion of a 220-question online survey. When compared to an adolescent Norwegian CL/P sample (Feragen et al., 2014), the reported rates of dyslexia and language impairment were higher in the UK CL/P cohort, but both samples remain in line with their respective general population reference groups. The rates of autism spectrum disorder, learning difficulties, and associated syndromes are lower in the UK adult cohort compared to the Norwegian CL/P sample. Given that Feragen and colleagues (2014) found the highest prevalence of additional conditions in those with a cleft palate only, these lower rates could be explained by the relative underrepresentation of cleft palate only in the present sample.

Furthermore, a considerable proportion of adults with CL/P were unaware as to whether they had a syndrome or not. It is likely that since participants were originally treated for CL/P, genetic testing has advanced considerably and become more integrated into CL/P services. Additionally, Feragen et al. (2014) reported a high rate of ‘suspected but undiagnosed’ cases of developmental delay. Routine screening is not currently carried out in all UK cleft teams (Stock et al., 2019), but should be a key consideration to ensure that children at risk of developmental problems are identified from an early age.

Individuals with comorbidities are often excluded from research samples, and the current study adds weight to previous literature suggesting that in doing so, critical knowledge on a vulnerable subgroup of patients could be lost (Feragen et al., 2014). It is therefore recommended that future studies document and categorise comorbid conditions wherever possible, to ensure these aspects are not neglected in research and that patients’ needs are being met in practice.

***Physical Health in Adulthood***

Participant responses indicated that the sample was in good physical health overall. Nonetheless, a number of challenges were evident, and were perceived to affect everyday life. In particular, breathing difficulties were prevalent, and many participants reported symptoms which suggest that breathing during sleep was suboptimal. Airway patency can be detrimentally impacted by CL/P as a result of restricted or blocked airflow through the nose, yet there has been little research on the long-term implications of this. Further, and despite the frequency of breathing-related concerns, only a small proportion of participants had ever received a breathing assessment. Investigating and addressing concerns related to breathing is not currently part of the UK cleft service specification (NHS England, 2013). It is therefore recommended that the results of the current study be further explored, and if replicated, that consideration be given to adopting a breathing assessment as part of routine CL/P care.

Similar findings were evident in regard to hearing difficulties. Although a high prevalence of hearing difficulties was identified in comparison to the general population, fewer than half of affected participants were receiving audiological care. Again, it is recommended that further consideration be given to the integration of routine hearing assessments, especially for adults returning to CL/P services later in life.

The majority of participants reported some degree of ongoing speech concerns and were not particularly content with their speech at the time of survey completion. In addition, although participants reported that their speech was usually understood by most people, they still perceived their speech to be noticeably different from that of others without CL/P. These findings reflect previous literature showing a relatively high incidence of functional speech impairments, including poor intelligibility and problems with resonance (Chuo et al., 2008; Gkantidis et al., 2015). Previous research has shown subjective dissatisfaction with speech to be associated with impaired social functioning and psychological risk in individuals with CL/P (Berger and Dalton, 2011; Havstam et al., 2011; Gkantidis et al., 2015), over and above objective ratings of speech quality as assessed by specialist speech and language therapists (Feragen et al., 2017). Furthermore, participants were not particularly satisfied with the outcomes of the SLT they had received growing up. This may reflect the UK cleft service pre-centralisation, at a time when speech outcomes were shown to be particularly poor (Sandy et al., 1998). A discussion with individuals about the perceived impact of their speech on everyday life and an exploration of potential interventions could therefore be an important part of all routine CL/P assessments, especially in the case of adults returning to the cleft service.

Although individuals born with CL/P do not typically have difficulties with eating or swallowing that would result in a diagnosis of dysphagia, few studies have investigated the more subtle behaviours that may be indicative of difficulties, such as avoidance of certain foods, or of social situations that involve eating or drinking. The findings of the current study suggest that although the majority of participants enjoyed eating and drinking and the social opportunities that it offers, a considerable proportion were experiencing unpleasant symptoms and/or making compromises on a daily basis. Although residual fistulae can be difficult to repair, exploration of alternative options, such as prosthesis to reduce the functional and social impact of fistulae on eating and drinking could be beneficial.

***Access to Specialist Healthcare***

Although not all participants had tried to access CL/P-related services via their GP, 44 participants had experienced difficulties with this. This was despite this group of participants being well-connected to primary healthcare services, with the vast majority having visited their GP at least once within the last year. Participants reported GPs to lack knowledge of CL/P and its treatment, to be dismissive of participants’ concerns, and/or to refer participants to the incorrect hospital or department. In some cases, participants also reported hospitals to be unaware of or reluctant to refer patients to specialist CL/P teams. These experiences were exacerbated in several cases by the participants themselves being unaware of their entitlement to specialist NHS services. Similar findings have been reported elsewhere, in the case of general dental practitioners (GDPs). For example, Searle et al. (2017) identified a lack of understanding of the issues surrounding CL/P among GDPs. Stock and colleagues (2018) also highlighted a lack knowledge of CL/P and its treatment among GDPs, as well as how and when to refer patients to specialist services. These authors suggested that improved training and resources, in addition to closer collaboration between specialist CL/P teams and local professionals was needed. Based on the findings of the current study, similar efforts may be needed here, in order to ensure adults are not made to wait an unreasonable amount of time, undergo private treatment at their own expense, or endure treatment which is ineffective or harmful. GP costs are increasing year on year in the UK (British Medical Association, 2015). Rather than being handled solely in the community, both GPs and patients could be better served if awareness among GPs was increased and the referral process was improved. Charitable organisations are also well-placed to empower adults with knowledge about the treatment they are entitled to, how to access it, and who to approach if they are struggling. Raising awareness among adults themselves could be particularly important given that participants were generally willing to explore further treatment options if this opportunity were made available to them.

***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. It would be beneficial for future studies to investigate trends for individuals with cleft palate only to better understand the experiences of this population. How to better represent adults who are less engaged with CL/P services and/or how to reach those who remain 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), 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. Additionally, similar studies undertaken in other countries would provide a useful comparison to measure the impact of cultural differences, and differences in healthcare model. Finally, the sample size of this study did not allow for analysis of results according to cleft type, gender, or other variables of interest. Previous studies have commented on the challenges of analysing subsets of data even when the overall sample is relatively large (e.g. Feragen et al., 2015). Multicentre, interdisciplinary, and international working 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**

Individuals with CL/P may be at risk of physical health issues persisting into adulthood. The monitoring of physical symptoms from an early age is recommended, as is the routine assessment of these symptoms in adults who return to the CL/P service later in life, particularly with regard to breathing, speech, hearing, and eating and drinking. Further, both GPs and patients themselves may be unaware of specialist CL/P services and how to access them. Multi-pronged education programmes, as well as improved links between primary and tertiary care settings are crucial for improving the referral process and preventing harm. Finally, it is recommended that future studies more thoroughly investigate the prevalence of comorbid conditions in CL/P, in order to address the knowledge gap regarding the longer-term health burden of the condition.

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