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

Abstract

Objectives: Previous research has suggested ~~that~~ adults ~~affected by~~with a cleft lip and/or palate (CL/P) may find their familial, friend, collegial, and romantic relationships ~~directly or indirectly~~ impacted by their condition. Building on this prior work, ~~the aim of the current~~this study ~~was to~~ examined the self-reported interpersonal experiences of a national sample of adults born with CL/P.

Design: An online survey was designed by the Cleft Lip and Palate Association in collaboration with (*University*). A total of ~~207~~ 181 eligible responses were collected between July and October 2018. Qualitative data were analysed using inductive content analysis, while quantitative data were analysed using descriptive statistics and independent t-tests.

Results: Most participants reported having good relationships with family members and existing friends with similar patterns of marriage and parenthood as the general population.~~,~~ ~~yet~~ Some experienced difficulties initiating new friendships and~~/or~~ romantic relationships. ~~Some participants~~Others described ~~instances of~~ bullying and~~/or~~ discrimination in adulthood, most often ~~occurring~~ in public settings and~~/or~~ the workplace. Participants reported lower satisfaction with intimate relationships and less secure attachment ~~styles~~ compared to the general population. Concerns regarding the heritability of CL/P and its impact on the decision to have a family were also ~~indicated~~reported.

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Conclusions: Individuals with CL/P are at risk of negative social experiences, which if not appropriately addressed may impact psychological wellbeing in adulthood. The findings illustrate the importance of ~~societal campaigns to raise awareness and target discrimination, in addition to~~ routine psychological support from childhood onwards to help individuals with CL/P build social skills, self-esteem, and social confidence, and to develop mutually fulfilling friendships and intimate relationships. Additionally, societal campaigns to raise awareness and target discrimination may be helpful.

Key words: cleft lip and palate; adult; psychological adjustment; social experiences; discrimination; romantic relationships

Introduction

Although the impact of cleft lip and/or palate (CL/P) on families and children is becoming increasingly understood, there is ~~only~~ limited research into the ongoing impact of CL/P in adulthood (Stock & Feragen, 2016). With a few exceptions, the majority of existing literature regarding adults with CL/P has ~~also~~ tended to focus ~~solely~~ on medical concerns. Studies that have explored adults' experiences more holistically have suggested that CL/P is a lifelong condition and that the concerns it poses in adulthood are multifaceted. For example, two large qualitative studies identified several areas of potential concern, including access to treatment, medical decision-making, social and romantic relationships, higher education and employment, and psychological health and support (Stock et al., 2015; Kappen et al., 2019). Additional research into the psychosocial wellbeing and support needs of adults with CL/P is essential for the delivery of lifelong patient-centred care.

Previous research has suggested that adults with CL/P may find their familial, friend, collegial, and romantic relationships to be impacted directly or indirectly by their diagnosis. For example, difficulties in engaging in social interaction and relationship formation, delayed emancipation from the family home, elevated levels of social anxiety, and low levels of social support have been identified (Cochrane & Slade, 1999; Berk et al., 2001). Further, individuals with CL/P may experience negative reactions to their appearance and~~/or~~ speech in social and~~/or~~ employment settings and may struggle to initiate romantic relationships (Patel and Ross, 2003; Stock et al., 2015; Feragen et al., 2016). These findings have been linked to social withdrawal (Cochrane & Slade, 1999), a lower rate of marriage and/or delayed marriage (Danino et al., 2005), and an overall impact on adults' social lives (Marcusson et al., 2001). Previous studies have also reported that adults with CL/P are more likely to be childless (Yttri et al., 2011) and~~/or~~ may find the prospect of starting their own family daunting, given the potential heritability of CL/P (O'Hanlon et al., 2012; Stock and Rumsey, 2015).

In recent years, the level of psychological support for individuals born with CL/P has increased in the United Kingdom (UK)~~,~~ as part of the reorganisation of cleft care delivered by the National Health Service (NHS; Sandy et al., 1998) and the growth of the leading UK charity, the Cleft Lip and Palate Association (CLAPA). In parallel, the volume of craniofacial literature focused on psychosocial aspects of care has increased~~,~~ and more insight into individuals' experiences of living with CL/P has been gained through qualitative exploration and subsequent recommendations for standardised outcome measurement (Stock, Feragen et al., 2018). Building on this recent work, the aim of the current study was to examine the self-reported interpersonal experiences of a national sample of adults born with CL/P~~,~~ via the administration of a comprehensive mixed-methods survey.

Method

Design

Drawing upon previous literature, ~~an online,~~a survey ~~design~~ was used by CLAPA in collaboration with (*University*) using the online survey platform~~,~~ SurveyMonkey to collect both quantitative and qualitative data. The survey was also available in paper format contained within a 54-page booklet. The survey consisted of 220 questions split across 12 sections. ~~Where possible, standardised measures were used.~~ An overview of survey content is 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). Given the volume of data collected from the survey, findings are reported across a series of papers, ~~of which~~including the current paper~~ is one~~. Interpersonal experiences were defined as participants' subjective views on the relationships that they have, or previously had, with family members, friends, romantic partners~~,~~ and members of the public.

Materials

The survey was designed using current literature (e.g. Stock & Feragen, 2016) and the expertise of the research team. Wherever possible, standardised measures were used. Where existing measures did not cover all topics of interest, questions were written by the research team. These 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”. [PLEASE ADD EXPLANATION OF WEIGHTED AVERAGES]

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).

Harter’s Self Perception Profile for Adults

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Self-perceived sociability and self-perceived competence in intimate relationships were evaluated using the Harter Self-Perception Profile for Adults (SPP-Ad; Sociability subscale; Intimate Relationships subscale; Messer & Harter, 1986). The Norwegian version of the scale was used, which is considered more straightforward to complete than the original and replicates the same factorial pattern, yet has been shown to achieve better reliability and convergent validity (Wichstrøm, 1995). All scale items were translated into English. Respondents were asked to indicate the degree to which they identify with a total of eight statements on a 4-point Likert scale (1 = describes me very poorly; 4 = describes me very well), where a higher score indicates a more competent self-judgement. A mean score was calculated and compared to a normative data sample consisting of 144 US adults aged 30-50 years (Messer & Harter, 1986).

Revised Adult Attachment Scale

The Revised Adult Attachment Scale (Collins, 1996) was used to assess participants' attachment styles in adult relationships. The scale contains three subscales, each composed of six items. The C~~LOSE~~lose subscale measures the extent to which a person is comfortable with closeness and intimacy. The D~~EPEND~~epend subscale measures the degree to which a person feels they can depend on others for support. The A~~NXIETY~~ nxiety subscale measures the extent to which a person is worried about being rejected or unloved. Participants were asked to respond to all subscales using a 5-point Likert scale (1 = not at all characteristic of me; 5 = very characteristic of me). High scores on the anxiety dimension characterise individuals who worry about being unloved or abandoned by romantic partners. High scores on the closeness dimension characterise individuals who find closeness with others easy, and high scores on the dependent dimension characterise individuals who feel that others are trustworthy and dependable (Collins, 1996). Mean scores were calculated for each subscale and compared to a normative sample of 68 'securely attached' US adults aged 18-25 (CITE).

Relationship Satisfaction Scale

The Relationship Satisfaction Scale (RS10; Røysamb et al., 2014) was used to evaluate participants' subjective satisfaction with their relationship with their current partner. Participants were asked to respond to ten items using a 6-point Likert scale (0 = strongly disagree; 5 = strongly agree). A higher score indicates a higher level of satisfaction. A mean score was calculated and compared to a normative sample of 347 Norwegian adults aged between 19-71 years (Røysamb et al., 2014).

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~~;~~, and 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, "How many close friends do you have? A. 0, B. 1, C. 2, D. 3, E. 4 or more; weighted averages, for example, "How much do you agree with the following statement? I have a good relationship with my mother currently [5 point scale from strongly disagree to strongly agree]"; as well as open ended questions, for example, "In your own words, describe why you believe your last relationship ended?" Where possible, data were compared to national statistics.~~ 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 second 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 second authors.~~ Descriptive statistics were used to analyse data collected from non-standardised 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.~~ Where possible, data were compared to national statistics. Scores obtained from standardised measures were compared to normative data and/or reference groups ~~where available~~

using independent samples t-tests. Cohen's *d* was used to calculate effect sizes, where values between 0.2 and 0.5 represent a small effect, values between 0.5 and 0.8 represent a medium effect, and values of more than 0.8 represent a large effect (Cohen, 1988).

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Results

Participants

A total of 224 responses were collected. Seventeen participants were excluded as they had been born outside of the UK (7.6%). Of the remaining 207 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 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 181 participants contributed data to 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).

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Familial Relationships

To gain a broad understanding of the varying dynamics in people's lives, participants were asked about their family background. ~~The majority~~Half of participants (*n* = 94; 53.1%) reported that their parents were currently either married or in a long-term relationship with each other. For most participants, neither of their parents had been born with CL/P *(n* = 160; 91.4%). Nine participants reported having a sibling born with CL/P (5.1%) and 39 reported that another family member, such as a cousin or grandparent had been born with CL/P (22.2%).

Participants were asked to respond to a series of statements and provide an answer on a five-point scale from Strongly Disagree (1) to Strongly Agree (5). Weighted averages were calculated as follows, listed from high to low: *"I have a good relationship with my mother currently"* (4.23); *"I had a good relationship with my mother when I was growing up"* (4.19); *"I enjoy spending time with my family"* (4.08); *"I had a good relationship with my father when I was growing up* (4.03); *"I have a good relationship with my father currently"* (3.99); *"I have a good relationship with my brothers and sisters currently"* (3.85); *"I know that I can rely on my family to provide me with emotional support during difficult times"* (3.85); *"I had a good relationship with my brothers and sisters during my childhood"* (3.66); *"I think of my childhood as a happy time"* (3.66); and "*I feel comfortable speaking with my family about my cleft"* (3.64).

Friendships and Sociability

The majority of the sample reported having four or more close friends (*n* = 106; 60.2%), and most rated these friendships as either 'excellent' (*n* = 87; 49.9%) or 'good' (*n* = 68; 38.6%). Participants were most likely to have made these close friends at work (*n* = 87, 50.3%) or secondary school (*n* = 78, 45.1%). University (*n* = 56, 32.4%), primary school (*n* = 46, 26.6%), and hobby groups (*n* = 43, 24.9%) were other effective places to develop close friendships.

Participants were asked to respond to a series of questions relating to the quality of their friendships and provide answers on a five-point scale from Strongly Disagree (1) to Strongly Agree (5). Weighted averages were calculated as follows, listed from high to low: *"I am supportive to my* *friends"* (4.39); *"My friends accept me for who I am"* (4.38); *"I am happy when I am with my friends"* (4.23); *"I have known my best friend(s) for a long time"* (4.15); *"I know that I can rely on my friends to provide me with emotional support during difficult times"* (3.99); *"My friendships are mutually beneficial"* (3.89); *"I feel comfortable speaking with my friends about my cleft"* (3.53); *"I regularly spend time with my friends"* (3.45); *"I tend to initiate contact with my friends more often than they initiate contact with me"* (3.02); *"I make new friends easily"* (2.97); and *"I often have arguments or fall out with my friends"* (1.80).

Perceived Sociability was assessed using the Social Competence subscale of the Harter SPP-Ad. Results are shown in Table 3. Adults with CL/P (M = 2.73 ± 0.69) reported significantly lower self-perceived social competence compared to the general population sample (M = 3.19 ± 0.61). Adults with CL/P generally agreed with about two thirds of positive self-statements. Calculation of Cohen's *d* indicated a medium effect (*d* = 0.71).

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Peer Support

~~The majority of~~Most participants stated they had previously spoken with someone else who was born with CL/P (*n* = 105; 63.3%). When asked on a five-point scale from Strongly Disagree (1) to Strongly Agree (5), participants reported an average of 4.22 for the statement *"Peer support through organisations such as CLAPA is important for improving mental wellbeing."*~~.~~

Bullying and Discrimination

Almost eighty-five percent of respondents (*n* = 153; 84.5%) indicated that they had experienced some form of bullying during their lifetime. As an adult, 36 participants (23.8%) reported having been bullied in the workplace, while 18 (11.9%) had experienced bullying online. A small percentage of participants (*n* = 10; 6.7%) stated they had been bullied by a romantic partner. Two female respondents (1.4%) disclosed behaviour against them which the authors would identify as acts of sexual assault.

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Thirty-nine participants (21.8%) reported that they had experienced discrimination. This included being mocked for their appearance or speech (*n* = 25; 64.1%), not being presented with equal opportunities (*n* = 20; 51.3%), not being considered for a job (*n* = 19; 48.7%) or a promotion (*n* = 7; 18.0%), and being discriminated against by a date or romantic partner (*n* = 15; 38.5%).

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Regardless of when the perceived bullying and/or discrimination took place, participants qualitatively stated that these negative social experiences had long-term effects. These most frequently included: a loss/reduction of self-confidence (*n* = 38), low self-esteem/self-worth (*n* = 35), feeling shy, introverted, lonely, or withdrawn (*n* = 28), increased anxiety (*n* = 19), a reduced trust in others (*n* = 12), feeling upset or unhappy (*n* = 12), and depression (*n* = 11). A minority of participants also reported anger (*n* = 8), a lack of career progression (*n* = 5), a negative impact on overall psychological wellbeing (*n* = 4), attempted suicide/suicide ideation (*n* = 4), development of an eating disorder (*n* = 4), shame (*n* = 4), a negative impact on the formation of romantic relationships (*n* = 3~~);~~), self-harm (*n* = 3), an inability to work (*n* = 2), feeling odd or different (*n* = 2), an inclination to bully others (*n* = 1), and homicidal ideation (*n* = 1). Twenty-six participants also stated that although these experiences had been difficult at the time, they had helped them to become more resilient and develop helpful coping mechanisms in the longer term.

Overall, when reporting their experience on a five-point scale, participants reported that they felt they were as safe and equitably treated as anybody else in their community and generally felt comfortable visiting public places. However, participants frequently reported that the government could do more to ~~tackle~~ address discrimination in the UK (*n* = 107; 60.1%).

Romantic Relationships

The relationship status of the participants is presented in Table 2. The most common way participants met their partner was through a friend (*n* = 24; 28.3%), although meeting in the workplace was also common (*n* = 23; 19.2%). Online dating and dating apps were the third most

Commented [JA8]: Consistency in % calculations common way to meet a partner (*n* = 21; 17.5%). Thirty-nine respondents (32.2%) reported that their relationship had started as a platonic friendship and had evolved into a relationship over time.

Participants were asked to respond to a series of statements pertaining to their views on relationships on a five-point scale from Strongly Disagree (1) to Strongly Agree (5). Weighted averages were calculated as follows, listed from high to low: *"I worry/have worried that I will never find someone"* (3.53); *"I seek out people that I feel will accept me"* (3.32); "*I feel that I lack confidence and experience when it comes to dating"* (3.21); *"I feel naïve and inexperienced compared to the people I'm dating/potential dates in my age group"* (3.09); *"I have not invited someone I like on a date as I have found this to be too difficult"* (3.08); *"I find the dating scene confusing"* (3.06); *"I don't know where to find like-minded people"* (2.89); "*I feel that I am lagging behind my age group when it comes to dating"* (2.84); *"I have entered into and/or stayed in a bad relationship because I feel/felt that I can't do any better"* (2.64); *"I struggle with and/or worry about kissing because I was born with a cleft"* (2.63); *"I have put/am putting a lot of effort into dating"* (2.55); and *"I would prefer to remain single"* (2.04).

Self-perceived competence in intimate relationships was assessed using the Intimate Relationships subscale of the Harter SPP-Ad. Results are shown in Table 3. Adults with CL/P (M = 2.82 ± 0.70) reported significantly lower self-perceived competence in intimate relationships compared to the general population sample (M = 3.12 ± 0.67). Responses from adults with CL/P were generally similar to the comparison sample. Calculation of Cohen's *d* indicated a small effect (*d* = 0.44).

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Participants were also asked to indicate in a free-text box why they felt previous relationships had ended. The ~~vast~~ majority (*n* = 94; 75.6%) gave responses that could be considered common irrespective of having a condition such as CL/P (e.g. infidelity). Other reasons were attributed by a minority of participants as directly or indirectly related to having CL/P, as follows: low self-esteem (*n* = 10), a lack of self-confidence (*n* = 3), a reduced trust in others (*n* = 2), speech difficulties (*n* =

1), and a lack of partner understanding of CL/P-related challenges (*n* = 1). Ten participants also reported emotional and/or physical abuse as the primary reason for ending a relationship.

Adult Attachment Styles

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The RAAS ~~(Collin, 1996)~~ was used to assess participants' attachment styles in adult relationships. Results are shown in Table 3. Compared to a sample of young adults categorised as having a 'secure' attachment, adults with CL/P reported significantly less favourable scores on all three subscales. Calculation of Cohen's *d* indicated large effects in relation to the ~~CLOSE~~ Close and ~~DEPEND~~ Depend subscales, and a small effect in relation to the ~~ANXIETY~~ Anxiety subscale.

Relationship Satisfaction

The RS10 ~~(Røysamb et al., 2014)~~ was used to evaluate participants' satisfaction with their current relationship. A total of 133 participants reported being in a relationship at the time of survey completion (see Table 2). Of these, 121 participants completed the RS10. Results are shown in Table 3. Compared to a sample of 347 adults aged 19-71 years, adults with CL/P reported significantly lower relationship satisfaction. Calculation of Cohen's *d* indicated a large effect.

Starting a Family

Brief information about the heritability of CL/P was provided in the survey. The majority of participants (*n* = 154; 90.1%) reported that they were already aware that CL/P could be inherited, while 17 (9.9%) stated that they learned this information for the first time from completing the survey. Of those participants who reported having biological children (*n* = 78; 45.9%), 48 (24.1%) stated that one or more of their children had been born with CL/P. Additionally, 55 participants (69.6%) reported that the possibility of having a child be born with CL/P had caused them concern while trying for a baby and/or during the pregnancy.

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The 48 participants who reported having a child born with CL/P were asked to what extent they agreed with a series of statements on a five-point scale from Strongly Disagree (1) to Strongly

Agree (5). Weighted averages were calculated as follows, listed from high to low: *"When my child was born with a cleft I felt worried for my child"* (3.51); *"When my child was born with a cleft I felt upset"* (3.40); *"When my child was born with a cleft I felt guilty"* (3.28); *"When my child was born with a cleft I felt shocked"* (3.09); *"Having a child has helped me to cope better with my own cleft-related concerns"* (2.98); *"My own experiences make it more difficult for me to cope with my child's cleft"* (2.80); *"When my child was born with a cleft I was blamed by others"* (2.55); and *"When my child was born with a cleft it was not a concern for me"* (2.48).

Participants were also asked their views on genetic testing. A total of 105 (63.6%) thought having this option was important as they wanted to be as informed as possible, while 26 (15.8%) stated they would rather not know and 14 (8.5%) felt the results would be too ambiguous to be helpful. The remaining 20 participants (12.1%) reported they had not given the topic any thought or had not been offered genetic testing.

Discussion

The aim of this study was to examine the self-reported interpersonal experiences of a national sample of adults born with CL/P. ~~To the authors' knowledge, this is the largest and most comprehensive survey of psychological adjustment in adults to date.~~ The findings provide valuable insight based on a large sample into the ongoing interpersonal challenges of individuals with CL/P once they reach adulthood. ~~Findings are discussed below in more detail and recommendations for clinical and community practice are provided.~~

Family and Friendships

On the whole, adults with CL/P who participated in the current study reported having positive relationships with family members and reported a high level of satisfaction with their existing friendships. In contrast to previous some quantitative work in this area stating that adults with CL/P have a low level of social support (Cochrane and Slade, 1999), adults in the present study felt their friendships were mutually beneficial, felt accepted by their friendship group, were able to talk with and rely upon their friends, and felt happy when in the company of friends. Social support is known to have positive benefits for general mental and physical health (Taylor, 2011), while perceived social acceptance has been shown to act as a buffer against depressive symptoms and appearance dissatisfaction in individuals with CL/P (Feragen et al., 2010). In addition, most participants reported having met another person born with CL/P. This is in contrast to other studies (e.g. Stock et al., 2015) and is possibly reflective of the recruitment strategy. ~~Nevertheless, A~~Aaccess to peer support was seen as an important aspect of psychological wellbeing.

Despite these positive findings, adults with CL/P reported ~~significantly~~ relatively lower self-perceived social competence compared to a normative US sample, indicating that they felt uncomfortable when meeting new people, questioned whether they were enjoyable to be with, and perceived themselves to be somewhat unsociable. A separate paper based on the same sample as the present paper (*authors*) identified elevated levels of fear of negative evaluation in adults with CL/P. Taken together, these findings suggest that although adults may feel comfortable in their existing friendships, engaging in new relationships may still be a cause of considerable concern.

Perceived Victimisation

Previous CL/P research has identified a link between social anxiety and perceived stigma and/or bullying (Alansari et al., 2014). In the present study, the majority of participants reported experiencing bullying at some time. While some participants felt these experiences had made them more resilient in the long term, others felt that the inverse was true. The most common themes to arise from these experiences included feelings of low self-esteem and self-worth, social anxiety, reduced confidence, and a reduction in trust of others. Although variable rates of teasing and bullying have been reported in CL/P samples to date (see Stock & Feragen, 2016), an association between these experiences and individuals' long-term psychosocial wellbeing has been well established (Lorot-Marchand et al., 2015; Feragen & Stock, 2016). There is also a risk that these difficult experiences may impact on future interactions with others and create a cycle of negative social interaction, loss of confidence, and withdrawal (Alansari et al., 2014).

A ~~novel and~~ concerning finding of the current study was that a significant minority also reported experiencing bullying and/or discrimination during adulthood. This was predominantly found to occur in public settings and in the workplace, with a minority of younger participants also reporting online bullying. Examples included a perceived lack of equal educational and vocational opportunities, being mocked for their appearance and/or speech, and being rejected by a potential romantic partner due to their condition. A recent report by the charity Changing Faces examined discrimination in individuals with appearance-altering conditions more generally (Changing Faces, 2017) and found a high prevalence of unwanted attention from strangers, incidences of mockery of appearance-altering conditions, and a longer-term impact of discrimination on individuals' educational and vocational aspirations. In a paper utilising the same sample of adults with CL/P as the current study, (*authors*) found a lower level of perceived job competence and satisfaction in comparison to norms. Although this negative impact on vocational experiences is not universally reported (Oosterkamp et al., 2007; Stock et al., 2015), the potential long-term effect of challenging social experiences and perceived discrimination should not be underestimated.

Intimate Relationships

In the present study, many participants reported concerns in relation to dating and some felt inexperienced in intimate relationships in comparison to their peers without CL/P. Some participants also expressed concerns about the impact of CL/P on functional aspects of intimacy, such as kissing. Concurrently, some adults' attempts to engage in the dating scene had been adversely affected, and on average participants reported significantly lower self-perceived competence in intimate relationships when compared to a normative group. Previous research has also identified adults with CL/P to experience difficulties in establishing romantic relationships

(Patel and Ross, 2003; Stock et al., 2015) and/or to engage in romantic relationships at a later stage in comparison to peers (Danino et al., 2005; Feragen et al., 2016). In the broader literature, avoidance of intimate relationships and a negative impact on sexual activity has been found in individuals with a range of appearance-altering conditions, such as skin conditions, facial birthmarks, scarring, and alopecia (Griffiths et al., 2012; Sharratt et al., 2018), as well as in those with poor body image more generally (Laus et al., 2018). Here, appearance may be considered as central to attraction~~,~~ and any perceived difference is therefore viewed as a barrier to the formation of romantic relationships (Sharratt et al., 2018). In turn, self-esteem may be affected~~,~~ and internal fears of being negatively judged or rejected may develop. Nonetheless, and contrary to some prior reports (e.g. Danino et al., 2005), the relationship status of the current sample did not seem to differ considerably from National Census data.

Participants in the present study were also asked to complete measures of adult attachment and relationship satisfaction, to evaluate the perceived quality of adults' intimate relationships once they had been established. When compared to normative samples, adults with CL/P described less secure attachment styles, suggesting that they felt more anxious in romantic relationships, found intimacy difficult, and were less trusting of their partner. Some adults reported having entered into unfavourable relationships because they felt they wouldn't be able to attract anyone more suitable. This is particularly concerning since a number of adults reported having experienced emotional, physical, and/or sexual abuse, which may be similar to the UK national rates of domestic abuse (cite). ~~No known prior studies have explored these topics in the context of adults with CL/P.~~ ~~However,~~ Sharratt and colleagues (2018) illustrated how adults with a range of appearance-altering conditions deemed themselves to be devalued by their different appearance. In several cases, these perceived deficiencies had led adults to try to compensate by cultivating other socially desirable characteristics (such as relative youth), to feel "lucky" to be in a relationship, and/or to settle for unrewarding relationships, as identified in the current study. Sharratt et al. (2018) describe a process in which the negatively perceived ~~defective~~ nature of the individual's appearance shapes

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Commented [JA14]: Please provide context of general population their belief that they are inadequate and that others will reject or discriminate against them (Kent and Thompson, 2002). This can lead to feelings of shame and anxiety, in addition to cognitive distortions that may influence the way in which social experiences are interpreted (Kent and Thompson, 2002). For other adults participating in the study by Sharratt and colleagues (2018), acknowledging the impact of the condition on the relationship could be difficult and communication could therefore be hindered. This could represent a contributory factor in the development of longer-term relationship dissatisfaction, which in contrast to previous research (Danino et al., 2005) was found to be lower compared to general populations norms.

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Having Children

Previous studies have reported that adults with CL/P are less likely to have their own children (Yttri et al., 2011) and/or may find the prospect of starting their own family unnerving, given the increased chance of having a child with the same condition (O'Hanlon et al., 2012; Stock and Rumsey, 2015). Given the relative age of the participants in the current study, as well as the number of adults who were married or cohabiting at the time of survey completion (see Table 2), the number of participants with children did not seem to differ considerably from National Census data. Nonetheless, the majority of participants reported ~~having (had)~~ concerns about starting a family. Participants whose children had been born with CL/P recalled difficult emotions at the point of diagnosis, including worry, distress, shock, and guilt. In contrast to this finding, some studies have reported no difference in the views of parents with and without CL/P (Andrews-Casal et al., 1998; O'Hanlon et al., 2012), while others have demonstrated adults with CL/P to have positive views of having children irrespective of recurrence risk (Patel and Ross, 2003). Following a qualitative investigation, Stock and Rumsey (2015) suggested that the degree of concern about having a child with CL/P may be considerably influenced by the individual's own experiences of growing up with the condition~~,~~ and the extent to which they had adjusted positively to these experiences. In line with Stock and Rumsey (2015), the majority of participants in the current study

Commented [JA16]: Please include in Table 2 were in favour of genetic testing, but not all felt this approach would be helpful due to the likely ambiguity of results.

Recommendations for Practice

Participants in the current study reported some difficulties in establishing new friendships and romantic relationships. Further, a portion of the sample reported dissatisfaction with intimate relationships was reported, alongside insecure adult attachment~~ styles~~. Where not already implemented, clinicians working with young people with CL/P could consider conducting routine assessments of social integration and liaising closely with schools to identify those at risk and ensure that any social concerns are addressed as early as possible. For both young people currently receiving care and adults who are no longer engaged in routine treatment, specialist one-to-one psychological support, useful topics for general advice from charitable organisations might include: ~~on instigating~~ starting relationships, ~~and~~ coping with difficult social encounters, access to peer support, ~~and~~ evidence-based support for tackling stigma, addressing workplace bullying, and developing social confidence~~ may be beneficial~~. In order to reduce prejudice and discrimination on a societal level, charitable organisations may be ideally placed to deliver targeted campaigns and appeal to government, particularly if multiple organisations ~~band together~~collaborate in their efforts. Awareness campaigns to foster public understanding of appearance-altering conditions and speech difficulties are also important. Finally, education regarding the causes and correlates of CL/P could benefit the cleft community as a whole, in order to support patients to become experts in their own diagnosis, and in turn to improve coping and health status (von der Lippe et al., 2017).

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. Participants also appeared more likely to have a child with a cleft than would be expected and this parental perspective may have a wide reaching impact on how they interpret their experiences as an adult. 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 (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. Third, ~~and~~ in order to compare the emotional wellbeing of individuals with CL/P to the general population, normative and/or comparison samples are required. In some cases, these comparison groups may not be ideal, due to cultural variation and/or a mismatch in key demographic factors such as age and gender. ~~However, the present study utilised normative populations that reported similar characteristics to the study sample as far as possible.~~ Statistical significance should also be interpreted within the context of reported effect sizes. Fourth, although participants indicated that difficult social experiences had helped them to be more resilient in the long-term, the current study did not include standardised measures of resilience ~~due to a limited budget and the need to keep the survey to a certain length~~. Work in the field of craniofacial conditions has identified a range of positive outcomes in addition to the known challenges (e.g. Eiserman, 2001). Although on average samples may produce unfavourable scores when compared to the general population, a subset of adults may still be coping well (Stock et al., 2015). Measures of positive aspects would therefore be important to include in future work in order to ensure a balanced perspective. 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. In addition, several standardised outcome measures were utilised. Consistency in the use of outcome measures is of key importance to the progression of knowledge, and the current study provides information on the utility of these measures for use with the adult CL/P population. 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 negative social experiences, which if not appropriately addressed may impact psychological wellbeing in adulthood. The findings illustrate the importance of routine psychological support from childhood onwards to help individuals develop social skills, self-esteem, and social confidence. Charitable organisations are also well placed to host awareness-raising campaigns and lobby government. The integration of such support could be effective in countering any unwanted attention, bullying, and discrimination~~,~~ and helping adults with CL/P to develop mutually fulfilling friendships and intimate relationships.

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