

Risk and protective factors for psychological distress in families following a diagnosis of cleft lip and/or palate

Abstract

Objectives: Despite the potential psychological impact of a diagnosis of cleft lip and/or palate (CL/P) on parents, few large-scale studies currently exist. Utilising data extracted from The Cleft Collective Birth Cohort Study, the current study aimed to examine the psychological impact of the diagnosis on parent and family functioning, and to identify risk and/or protective factors contributing to parental adjustment in order to inform future psychological intervention.

Methods: Parent-reported questionnaire data were extracted for 1,163 parents (644 mothers and 519 fathers). Outcome measures included the PedsQL-Family Impact Module, the Perceived Stress Scale, the Hospital Anxiety and Depression Scale, and a condition-specific tool designed by the Psychology Clinical Excellence Network.

Results: Overall, findings suggest that parents adjust well to the diagnosis. Factors found to be protective against psychological distress for both mothers and fathers included a positive life orientation, satisfaction with healthcare, and relationship satisfaction. Close friendships were also protective against depression in mothers. Risk factors for mothers included the presence of a prior mental health condition, and stressful life events during pregnancy. Risk factors for fathers included being older at the time of conception, and recently being absent from work.

Conclusions: Findings suggest a need for appropriate psychological screening of both parents following a diagnosis of CL/P and emphasise the importance of coordinated multidisciplinary care for psychological health. Preventative models of intervention to strengthen familial relationships and build resilience require further investigation.

Keywords: cleft lip and palate; cohort study; parental wellbeing; family resilience; The Cleft Collective

Introduction

A diagnosis of cleft lip and/or palate (CL/P) can invoke feelings of shock, guilt, and grief in parents, as well as concern for their child's future (Nelson, Glenny et al., 2012). Parents must come to terms with the implications of their child's condition, process a wealth of new information, grapple with feeding difficulties, and manage potentially uncomfortable reactions of friends, family members, and members of the public (Nelson, Glenny et al., 2012). Preparation for surgery has been described as a particularly distressing time for parents, who must deal with conflicting emotions surrounding the sanctioning of surgical intervention on their new-born (Nelson, Kirk et al., 2012). Given the potential impact of CL/P on parental wellbeing and the known implications of poor parental adjustment on children's later development in the broader literature (Sanger et al., 2015), the provision of evidence-based psychological support for families during periods of difficulty is a crucial priority.

Psychology is a relatively new discipline in the field of CL/P, having been introduced as a recommended component of CL/P teams in the UK following the centralisation of cleft care in the early 2000s (Sandy et al., 1998). As a result, understanding of how families adjust to a diagnosis of CL/P and its associated treatment remains limited, and little evidence for psychological intervention currently exists (Norman et al., 2015). Although recent qualitative work has offered in-depth reports of families' experiences of the treatment journey (e.g. Nelson & Kirk, 2013; Nelson, Kirk et al., 2012; Stock & Rumsey, 2015; Vanz & Ribeiro, 2011), the quantitative literature is characterised by conflicting findings. While some studies have observed high levels of parental stress and depression (e.g. Habersaat et al., 2018), others have reported few differences in family functioning between parents of children with CL/P and the general population (e.g. Crerand et al., 2015).

Such discrepancies in the literature can be attributed largely to methodological limitations. Primarily, these include a lack of large/multicentre samples and inconsistencies in outcome measurement (Stock, Feragen et al., 2018). Although common to many areas of health research and

challenging to overcome, these limitations are detrimental to the rate of improved understanding and to the transfer of knowledge to clinical practice. In addition, most previous research has focused on the experience of mothers, and thus little is understood about the psychological impact on fathers, or the family unit as a whole (Stock & Rumsey, 2015; Zeytinoglu et al., 2016a). Crucially, few studies have investigated the potential risk and/or protective factors which may contribute to individual differences in adjustment (Stock & Feragen, 2016). An understanding of these factors is crucial for the early identification of those families at risk and the prevention of long-term psychological distress.

In order to tackle these known methodological challenges and to address the unanswered questions important to all stakeholders (James Lind Alliance, 2012), a UK-wide programme of research, entitled 'The Cleft Collective' was established in 2012 (Stock, Humphries et al., 2016). At the core of this programme sits a national birth cohort study, responsible for the collection of biological samples and longitudinal questionnaire data from the point of diagnosis onward. Members of all disciplines (plastic surgery, maxillofacial surgery, dentistry, orthodontics, speech and language therapy, audiology, ear nose and throat. genetics, paediatrics, nursing, and psychology) from all 16 specialist CL/P sites contributed to the design and implementation of the study, in addition to patient and parent representatives recruited predominantly through the leading UK CL/P charity, the Cleft Lip and Palate Association. All UK families receiving a diagnosis within the study period are eligible to participate, and data have been successfully collected from participating families since December 2013.

Utilising cross-sectional baseline questionnaire data extracted from The Cleft Collective Birth Cohort Study, the aim of the present study was to examine parental psychological adjustment following a diagnosis of CL/P in new-borns. Specifically, this study aimed to address two research questions: 1) What is the psychological impact of a diagnosis of CL/P on the family? 2) What factors are associated with parental psychological distress and/or adjustment?

Methods

Procedure

Ethical approval to establish The Cleft Collective Cohort Studies was granted by the South West Central Bristol Ethics Committee. Global Research and Development (R&D) approval was provided by University Hospitals Bristol. Local R&D approvals were subsequently obtained from each National Health Service (NHS) Trust. Parents (biological mothers and their partners) were approached to participate in The Cleft Collective Birth Cohort Study by a Research Nurse following referral to their local NHS cleft team. Parents were given verbal and written information about what participation in the cohort study would entail, and essential ethical details including their right to confidentiality and their right to withdraw. Hand-written informed consent was then obtained from every participating member of the family. Parents were specifically asked for permission to use their data in the future for individual ethically approved research studies. Participants completed The Cleft Collective baseline questionnaire pack in the period between the birth of their child and their child's primary surgery and returned their data anonymously via post to The Cleft Collective team at the University of Bristol.

Institutional ethical approval to analyse a subset of the data for the purpose of the present study was obtained from the (*Faculty*) Research Ethics Committee at (*University*). Confidentiality agreements to access the data were signed by the authors, and data were subsequently de-identified and transferred to the authors in a password-protected file.

Outcome Measures

Historically, consensus regarding key psychological constructs and outcome measures in the field of CL/P has been difficult to achieve (Strauss & Broder, 1991; Stock, Feragen et al., 2018). The UK Craniofacial Psychology Research Subgroup, consisting of Cleft Specialist Clinical Psychologists and Research Psychologists, was established in 2012 with the aim of coordinating clinically relevant research activity. First, a conceptual framework was designed and evaluated

with the support of the wider clinical community and Public Involvement representatives (Stock, Hammond et al., 2016). Corresponding standardised outcome measures were then comprehensively assessed according to their potential scientific contribution, their clinical utility, and several pragmatic considerations. Patient and parent feedback was utilised to further refine the measures and ensure acceptability to participants.

The outcome measures utilised in the present study included the following: *The Pediatric Quality of Life Inventory – Family Impact Module* (PedsQL-FIM; Varni, Sherman et al., 2004) is a 36-item parent-reported measure of the impact of the child’s health on the parent’s Physical, Emotional, Social, and Cognitive Functioning, their Communication with others, and their Worry for their child, in addition to the impact on the families’ Daily Activities and Relationships. Items are rated on a 5-point Likert scale (0 = Never; 4 = Almost Always) and a higher score indicates better functioning. A total score, Health-Related Quality of Life summary score, and Family Functioning summary score are calculated, in addition to the eight subscale scores. *The Perceived Stress Scale* (PSS-10; Cohen et al., 1983), is a 10-item self-reported measure of perceived stress during the past month. The measure reflects the degree to which situations in the person’s life are appraised as stressful. Items are rated on a 5-point Likert scale (0 = Never; 4 = Very Often) and a higher score indicates a higher level of perceived stress. *The Hospital Anxiety and Depression Scale* (HADS; Zigmond & Snaith, 1983) is a 14-item self-reported measure of common ‘symptoms’ related to anxiety and depression during the past month. The measure consists of seven questions associated with anxiety (HADS-A) and seven questions associated with depression (HADS-D), rated on a 4-point Likert scale (e.g. 0 = Not at All; 3 = Most of the Time). Higher scores indicate a higher level of emotional distress, whereby a score of 0-7 is considered ‘normal’, 8-10 is considered ‘borderline’, and 11+ is considered to be clinically concerning. *The Clinical Excellence Network Questionnaire* (CEN-Q; baseline version) is a 7-item self-reported condition-specific measure (Stock, Hammond et al., 2016). The measure reflects the degree to which parents appraise their child’s health condition as stressful or threatening. The CEN-Q is a non-validated instrument,

which was specifically designed in accordance with existing literature, clinical input, and Public Involvement (Stock, Hammond et al., 2016). Items are rated on a 5-point Likert scale (0 = Never; 4 = Almost Always) and a higher score indicates more psychological distress.

Predictor Variables

Three standardised outcome measures were used as potential predictor variables: The *Pediatric Quality of Life Inventory – Healthcare Satisfaction Generic Module* (PedsQL-HSGM; Varni, Burwinkle et al., 2004) is a 24-item parent-reported measure assessing six dimensions of healthcare satisfaction (Information, Inclusion of Family, Communication, Technical Skills, Emotional Needs, and Overall Satisfaction). Items are rated on a 5-point Likert scale (0 = Never; 4 = Almost Always) and a higher score indicates greater satisfaction. The *Revised Life Orientation Scale* (LOT-R; Scheier et al., 1994) is a 10-item measure of optimism and pessimism. Items are rated on a 5-point Likert scale (0 = Strongly Disagree; 4 = Strongly Agree) and a higher score indicates a more positive life orientation. The *Social Readjustment Rating Scale* (SRRS; Holmes & Rahe, 1967) is a 43-item measure of stressful life events occurring in the last year. Each event on the scale is assigned a value (11-100), which are later summed to obtain an overall score. A score of 300 or more indicates a high risk of illness, a score of 150-299 indicates a moderate risk, and a score of less than 150 indicates a mild risk. The *Relationship Satisfaction Scale* (RS10; Røysamb et al., 2014) is a 10-item measure of an individual's subjective satisfaction with their relationship with their current partner. Participants respond using a 6-point Likert scale (0 = Strongly Disagree; 5 = Strongly Agree) and a lower score indicates a higher level of satisfaction.

Additional bio-demographic data (such as cleft type and family health history), and single-item psychological data (such as satisfaction with close friendships) were derived from Cleft Collective questionnaires and families' medical notes. A full list of included variables is available as supplementary material. A data dictionary detailing the variables collected in The Cleft Collective Birth Cohort Study is also available at: www.bristol.ac.uk/cleft-collective/professionals/access.

Analysis

A review, verification and validation of the database was undertaken prior to descriptive and inferential analysis. There were no unduly large or strongly influential observations in the sample. Given the relatively large sample sizes, reliance can be placed on the result of the Central Limit Theorem, and parametric statistical tests were therefore performed. These tests included Pearson's correlation coefficient (r) as an index of strength between two scale variables. For statistically significant correlations, absolute r values of approximately 0.1 in magnitude are considered to represent a small effect, 0.3 to represent a medium effect, and 0.5 to represent a large effect (Cohen, 1988). The paired samples t -test was used to compare parent dyads on each outcome measure, and the effect size was calculated using Cohen's d . For tests of difference, values for d 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). The robust one-way analysis of variance (ANOVA) was used to assess the relationship between the scale outcome variable and each categorical variable. Prior to conducting the regression analyses, a series of exploratory analyses were performed in order to determine which variables were eligible for inclusion in the models. These preliminary analyses are available as supplementary material. Analyses included Pearson correlation (when both the DV and the IV were continuous), the robust one-way ANOVA (when the DV was continuous and the IV was categorical with more than two categories), or the Welch version of the independent samples t -test (when the DV was continuous and the IV was categorical with only two categories). Based on the outcome of these tests, variables were included in the regression models if they met the following conditions: a) sample n was large enough so as not to significantly affect overall sample size (Brooks & Barcikowski, 2012); b) the variable was associated with two or more outcomes at univariate analyses stage; and c) inclusion of a variable did not cause multicollinearity problems (i.e. all variance inflation factors are less than 4).

Norm scores for the PedsQL-FIM and the PSS-10 were derived from Medrano and colleagues (2013; USA), and Cohen (1988; USA) respectively. Clinical cut-off scores were used to interpret

scores obtained using the HADS, in addition to normative data derived from Crawford and colleagues (Crawford et al., 2001; UK). The CEN-Q is a novel measure, and therefore further analyses were conducted to examine response distribution. The extent of agreement between mother and father dyads on items of the CEN-Q was measured using Cohen's Kappa, the strength of correlation using the Goodman-Kruskal gamma statistic, and the degree of systematic differences was measured using the McNemar-Bowker statistic.

Results

Participants

Participants in this study comprised 1,163 parents of children born with CL/P, who contributed baseline questionnaire data to The Cleft Collective Birth Cohort Study between December 2013 and December 2017. This included 644 mothers and 519 fathers, with 497 parent dyads. Participant characteristics are provided in Table 1. In comparison to UK Census data (Office for National Statistics, 2018), the sample was found to be a predominantly White, UK-born, educated population. Participants also reported above average median household income for two parent families (Office for National Statistics, 2018). When considering unregistered data, the distribution of the child's cleft type in the current sample was found to be relatively comparable with the national data reported by the Cleft Registry and Audit Network in 2017.

Mean Scores and Associations

To address the first research question, mean scores for each of the five outcome variables were calculated for mothers and fathers (Table 2). Mothers scored significantly less favourably than fathers on all outcome measures, except the PedsQL-FIM Family Functioning summary score. Effect sizes were small to medium (Table 2). Associations between scores on outcome measures were calculated for the 497 parent dyads. Mothers' and fathers' scores on all outcome measures were significantly positively correlated ($r = .223-.448$). These associations, in combination with

findings from the exploratory analyses (supplementary material), suggest that separate regression modelling for mothers and fathers is required.

Comparisons to Normative Data

To address the first research question, Mothers' and fathers' scores were compared to published norms (Table 3). Mothers scored significantly less favourably than norms on the Worry and Communication subscales of the PedsQL-FIM (medium effect size), and reported significantly higher levels of anxiety and depression (small effect size) and perceived stress (large effect size) compared to norms. In contrast, mothers reported significantly more favourable scores compared to norms on the total (small effect size), and Family Functioning (medium effect size) and Health-Related Quality of Life summary (small effect size) scores of the PedsQL-FIM, and the Emotional, Daily Activities (small effect size), and Family Relationships (large effect size) subscales of the PedsQL-FIM. Mothers also reported a less favourable score compared to norms on the Cognitive Functioning (small effect size) subscale of the PedsQL-FIM, and more favourable scores on the Physical Functioning and Social Functioning (small effect size) subscales of the PedsQL-FIM, but these differences were not found to be statistically significant. Fathers reported significantly higher levels of depression (small effect size) and perceived stress (large effect size) compared to norms. In contrast, fathers scored significantly more favourably than norms in relation to all of the scales of the PedsQL-FIM (small to large effect size) and reported significantly lower anxiety (small effect size) scores compared to norms. Both mothers' and fathers' scores on the HADS were found to be within the 'normal' range.

Regression Models - Mothers

To address the second research question, regression models were calculated for mothers. Following preliminary analysis of the data (supplementary material), the following variables were included in the regression models for mothers: PedsQL-HSGM, LOT-R, SRRS, relationship satisfaction, current household situation ("number of people who live with you"), number of close friends,

satisfaction with close friendships (rated on a 4-point Likert scale from Poor to Excellent), a prior mental health condition(s), illness during pregnancy, and child's cleft type. A summary of the regression models for mothers is shown in Table 4.

PedsQL-FIM: The fitted model accounted for 30.3% of the variance and comprised four statistically significant variables (adjusted $R^2=.303$, $F(10,361)=17.160$, $p<.001$). The existence of a prior mental health condition(s) and lower relationship satisfaction were negatively associated with mothers' quality of life scores, while a positive life orientation and greater healthcare satisfaction were associated with better functioning.

PSS-10: The fitted model accounted for 11.2% of the variance and comprised four statistically significant variables (adjusted $R^2=.112$, $F(10,347)=5.514$, $p<.001$). Greater healthcare satisfaction and a positive life orientation were negatively associated with mothers' perceived stress scores, while previous stressful life events and the existence of a prior mental health condition(s) were associated with higher levels of stress.

HADS-A: The fitted model accounted for 29.8% of the variance and comprised four statistically significant variables (adjusted $R^2=.298$, $F(10,347)=16.180$, $p<.001$). A positive life orientation and greater healthcare satisfaction were negatively associated with mothers' anxiety scores, while lower relationship satisfaction, and the existence of a prior mental health condition(s) were associated with higher levels of anxiety.

HADS-D: The fitted model accounted for 27.4% of the variance and comprised four statistically significant variables (adjusted $R^2=.274$, $F(10,348)=14.535$, $p<.001$). A positive life orientation, greater healthcare satisfaction and positive relationships with close friends were negatively associated with mothers' depression scores, while lower relationship satisfaction was positively associated.

CEN-Q: The fitted model accounted for 11.9% of the variance and comprised two statistically significant variables (adjusted $R^2=.119$, $F(10,350)=7.794$, $p<.001$). A positive life orientation and greater healthcare satisfaction were negatively associated with mothers' CEN-Q scores.

Regression Models - Fathers

To address the second research question, regression models were calculated for fathers. Following preliminary analysis of the data (supplementary material), the following variables were included in the regression models for fathers: PedsQL-HSGM, LOT-R, SRRS, relationship satisfaction, age at conception, an unexpected work absence, satisfaction with close friendships (rated on a 4-point Likert scale from Poor to Excellent), and satisfaction with employment ("I enjoy my work" rated on a 4-point Likert scale from Disagree to Agree). A summary of the regression models for fathers is provided in Table 5.

PedsQL-FIM: The fitted model accounted for 20.7% of the variance and comprised three statistically significant variables (adjusted $R^2=.207$, $F(7,396)=16.014$, $p<.001$). Lower relationship satisfaction was negatively associated with fathers' PedsQL-FIM scores, while a positive life orientation and greater healthcare satisfaction were associated with better functioning.

PSS-10: The fitted model accounted for 3.8% of the variance and comprised one statistically significant variable (adjusted $R^2=.038$, $F(7,388)=3.229$, $p<.01$). Older age at conception was positively associated with fathers' perceived stress scores.

HADS-A: The fitted model accounted for 20.0% of the variance and comprised three statistically significant variables (adjusted $R^2=.200$, $F(7,379)=14.796$, $p<.001$). A positive life orientation was negatively associated with fathers' anxiety scores, while an unexpected absence from work and lower relationship satisfaction were associated with higher levels of anxiety.

HADS-D: The fitted model accounted for 19.6% of the variance and comprised four statistically significant variables (adjusted $R^2=.196$, $F(7,383)=14.576$, $p<.001$). A positive life orientation and

greater healthcare satisfaction were negatively associated with fathers' depression scores, while lower relationship satisfaction and older age at conception were positively associated.

CEN-Q: The fitted model accounted for 10.6% of the variance and comprised three statistically significant variables (adjusted $R^2=.106$, $F(7,385)=7.614$, $p<.001$). A positive life orientation and greater healthcare satisfaction were negatively associated with fathers' scores on the *CEN-Q*, while lower relationship satisfaction was positively associated.

Discussion

To the authors' knowledge, this is the largest study to date to examine parental psychological wellbeing in the postnatal period. The aim of the present study was to examine parental psychological adjustment following a diagnosis of CL/P by analysing baseline questionnaire data extracted from The Cleft Collective Birth Cohort Study. Specifically, this study aimed to address two research questions: 1) What is the psychological impact of a diagnosis of CL/P on the family? 2) What factors are associated with parental psychological distress and/or adjustment? The findings provide important insights into psychological adjustment in this population and offer guidance for both clinical practice and future research.

Parental Psychological Adjustment Following a Diagnosis of CL/P

General Wellbeing

Previous research in the fields of CL/P and chronic illness has documented parents' emotional and social struggle to adjust to their child's condition and its long-term implications (Cousino & Hazen, 2013; Nelson, Glenny et al. 2012). These findings are also reflected in the present study, with mothers reporting higher levels of general anxiety and depression in comparison to norms, and both mothers and fathers reporting higher levels of perceived stress. In addition, analyses indicated that in comparison to norms, mothers were more likely to worry about the impact of the condition on the child and the family, and to struggle to communicate their concerns to others. Previous work by

Nelson, Kirk, and colleagues (2012) highlighted the conflicting emotions that parents of infants with CL/P experience, particularly in the time between diagnosis and primary surgery. Feelings of social exclusion and stigmatising reactions from others may be particularly distressing for mothers and may evoke anxiety for their child's future (Nelson, Kirk et al., 2012). Given that parental distress has the potential to impact considerably on attachment representations and longer-term child development (e.g. Pope et al., 2005; Murray et al., 2010), these findings add further emphasis to the need for psychological screening in the first few months following a diagnosis, to facilitate the identification of parents who are struggling and the provision of appropriate psychological support. Nonetheless, effect sizes varied from small to large across outcomes, and findings should therefore be interpreted with this in mind.

Overall, mothers reported less favourable scores on all but one of the measured outcomes when compared to fathers. Fathers have been somewhat neglected in past CL/P research (Nelson, Glennly et al., 2012), but when included, have reported fewer symptoms of psychological distress than mothers (e.g. Cole et al., 2016; Nidey et al., 2016). This is also reflected in the broader health literature (Cousino & Hazen, 2013). Nonetheless, qualitative studies have demonstrated that fathers report similar challenges to mothers in relation to their child's CL/P diagnosis and treatment and play a crucial role in the upbringing of their child (Stock & Rumsey, 2015; Zeytinoğlu et al., 2016a). Further, fathers may present a strong outward demeanour in support of their family following the initial CL/P diagnosis, and not share their own concerns until a later stage (Stock & Rumsey, 2015). Efforts should therefore be made to involve fathers in clinical care wherever possible and to follow fathers throughout the treatment pathway to ensure their needs are consistently met.

Family Resilience

In contrast, both mothers and fathers reported a higher level of functioning compared to norms on several measures, including overall health-related quality of life and family functioning. These findings are consistent with another large-scale study of family functioning (Cramer et al., 2015),

which found overall scores to be suggestive of parental adaptation, and healthy levels of family cohesion, expressiveness, and conflict. Recent work has also drawn attention to a range of positive outcomes among parents of children with CL/P, including heightened empathy for others, stronger familial relationships, and personal growth (Nelson, Glenny et al., 2012; Stock & Rumsey, 2015; Zeytinoglu et al., 2016a). The findings of the present study suggest that further exploration of the positive outcomes associated with CL/P is warranted.

While most studies in the broader literature have focused on the development of resilience in individuals, recent calls have been made to adopt a developmental systemic approach, in which the multilevel capacity of the family unit as a whole is considered (Walsh, 2016). Small scale qualitative studies have not only demonstrated the psychological impact of CL/P on wider family members, such as siblings and grandparents, but also the potential contribution that these family members can make to positive family adjustment (Stock, Stoneman et al., 2016, *manuscript in press*). Further exploration of the factors contributing to family resilience, and the interventions shown to be effective in the general population (Benzies & Mychasiuk, 2009; Dunst & Trivette, 2009), may support the application of related interventions to the field of CL/P.

Risk and Protective Factors for Psychological Distress

Regression analyses identified several factors that were consistently associated with psychological outcomes. These included: optimism, satisfaction with healthcare, relationship satisfaction, and several pre-existing factors.

Optimism

A positive life orientation was associated with almost all outcomes for both mothers and fathers, including lower levels of anxiety and depression, more positive appraisals of CL/P, and a lesser overall impact on the family. Additionally, optimism was protective against stress in mothers. The key role of optimism is well documented in the broader health literature and has been linked to more favourable ratings of subjective wellbeing in times of adversity, a higher use of constructive

coping strategies, better physical functioning, and more positive social experiences (Carver et al., 2010). Unfortunately, the construct of optimism, and its unique link to resilience, has been largely neglected in relation to CL/P. While a small number of studies have pointed to dispositional style as a potential protective factor among individuals affected by CL/P and their families (e.g. Baker et al., 2009; Sischo et al., 2016), few studies have explored this relationship in more depth, or sought to examine how an intervention to increase optimism among patients and their families may be beneficial. Given the view that optimism can be learned (Seligman, 1991), exploration of how optimism may feed into the delivery of psychological intervention at times of difficulty or transition, as well as the overall prevention of distress, is warranted.

Satisfaction with Healthcare

In the current study, healthcare satisfaction was associated with lower levels of depression, more positive appraisals of CL/P, and less of an impact on the family, as reported by both mothers and fathers. For mothers, healthcare satisfaction was also protective against anxiety and perceived stress. Previous research has emphasised the importance of healthcare delivery in providing reliable information and reducing psychological distress among parents of children born with CL/P (e.g. Knapke et al., 2010; Nelson & Kirk, 2013; Vanz & Ribeiro, 2011). In particular, parents of children with craniofacial conditions are thought to value health professionals' knowledge, technical competence, interpersonal skills, and dependability (Beaune et al., 2004; Johns et al., 2018). In the UK, cleft care was centralised in the early 2000s, following recommendations made by the Clinical Standards Advisory Group (Sandy et al., 1998). Several studies have since reported on the positive impact of these changes on several treatment outcomes, hospital episode statistics, and consistency in the national delivery of CL/P protocols (e.g. Fitzsimons et al., 2012; Scott et al., 2014; Waylen et al., 2015). Correspondingly, positive changes in the views of parents (Cleft Lip and Palate Association, 2007) and patients (Stock, Anwar et al., 2018) have also been reported. The findings of the present study lend further support to the importance of coordinated, multidisciplinary care for psychological health. Further dissection of the findings of the current

study could add to the literature by identifying the factors associated with healthcare satisfaction and highlighting which aspects of healthcare could be further improved.

Relationship Satisfaction

Mothers and fathers who were more satisfied with their relationship reported lower levels of anxiety, depression, and a lesser impact of their child's condition on family life. In addition, fathers who were more satisfied with their relationship were more likely to view their child's condition in a positive light. Further, associations between mothers' and fathers' scores on all outcome measures were found among parent dyads, indicating that mothers and fathers closely influence one another's wellbeing. These findings highlight the importance of familial relationships in the context of psychological adjustment to CL/P and point towards the potential benefits of a systemic approach to psychological intervention. Further examination of how couples manage the impact of having a child with a congenital craniofacial condition would be useful in future (Zeytinoglu et al., 2016b).

More broadly, social support is known to act as a buffer against the negative effects of acute and chronic stress in the general population and has been shown to be important in helping families to adjust to chronic illness (Baker et al., 2009; Cousino & Hazen, 2013). Although participants' satisfaction with close friendships was not found to be a pertinent variable in the current analyses, it was found to be protective against depressive symptoms in mothers. Cleft teams could help parents to identify close friends with whom they feel able to share their experiences and signpost parents to peer support services, such as those offered by charitable organisations.

Pre-Existing Factors

Additional factors predicting negative outcomes for mothers included the existence of a prior mental health condition (associated with higher levels of stress and anxiety, and a higher impact of the child's condition on family life), and recent stressful life events (associated with higher levels of stress). Additional factors predicting negative outcomes for fathers included being older at the time

of conception (associated with higher levels of stress and depression), and an unexpected absence from work (associated with higher levels of anxiety). While not amenable to change, these factors could still be screened for and monitored in practice.

Cleft Type

Cleft type was not found to be associated with any psychological outcomes in the present study. Contrary to the idea that a more objectively “severe” condition would have a greater impact on familial wellbeing, several studies in CL/P and related fields have consistently found cleft type/severity/visibility to be a poor predictor of overall psychological adjustment (Moss, 2005; Appearance Research Collective, 2009). Nonetheless, the treatment journey is likely to differ in some ways depending upon cleft type and any additional needs. Qualitative research may be best placed to examine these nuances.

Methodological Considerations

Representativeness of the Sample

Comprehensive data were extracted from The Cleft Collective Birth Cohort Study (Stock, Humphries et al., 2016). As such, participants had been recruited on a national scale, and eligibility criteria were highly inclusive. Nonetheless, the sample obtained for the purpose of the current study consisted predominantly of White, UK-born, educated families, with above-average income, with those from Black and Minority Ethnic communities, those having immigrated to the UK, and those with lower socioeconomic status, underrepresented in the current sample. Literature from a range of health fields has demonstrated clear differences in the way these groups interact with health services, and engage with research (Public Health England, 2017). Although not found in the present study, possibly due to a lack of sample diversity, several CL/P-focused studies have also been indicative of poorer outcomes among these subgroups (Stock & Feragen, 2016). Discussions with Research Nurses responsible for recruitment to The Cleft Collective Birth Cohort Study (Zucchelli et al., 2018) were suggestive of language as a barrier to informed consent, in addition to

the discomfort experienced by some nurses in approaching vulnerable families. While underrepresentation in study samples undoubtedly affects the overall conclusions of the research, it also results in little being learned about the experiences and support needs of potentially vulnerable groups. Further work is needed to ensure that studies are relevant and accessible to all eligible participants.

In the current study, data were collected via parent-reported questionnaires. Unfortunately, these were not always completed in full. Despite an overall sample of 1,163 participants, sample sizes were affected when taking missing data into account in the regression analyses. National studies take time to establish but are the field's best opportunity to collect large and representative datasets. Future efforts to streamline questionnaires, and to make questionnaires available online may help to facilitate more complete data collection (Zucchelli et al., 2018).

Utility of Measures

In efforts to lay the groundwork for a consensus in relation to key psychological constructs and outcome measures in the field of CL/P (see Stock, Feragen et al., 2018), the measures used in the current study were selected following a comprehensive evaluation process (Stock, Hammond, et al., 2016). Particular care was taken to include both 'generic' and condition-specific constructs. Based on the findings of the current study, the chosen measures appear to provide scientifically and clinically relevant data. Consistency in the use of outcome measures is of key importance to the progression of knowledge, and these measures could therefore be prioritised for use in future research. The utility of these measures to identify those at risk of psychological distress could also be tested further in the context of clinical assessments.

Conclusions

This study analysed baseline questionnaire data collected from 1,163 parents of infants born with CL/P. Taken together, the findings suggest that despite the psychological strain of having a child born with this long-term condition, most mothers and fathers adjust well overall. This may also

reflect the important contribution of the Clinical Nurse Specialists and the Clinical Psychologists embedded within UK CL/P teams. Nonetheless, findings suggest a need for appropriate psychological screening of both parents following a diagnosis of CL/P and emphasise the importance of coordinated multidisciplinary care for psychological health. Preventative models of intervention to strengthen familial relationships and build resilience require further investigation.

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