The Cleft Palate-Craniofacial Journal

Satisfaction with Healthcare in Families Following a Diagnosis of Cleft Lip and/or Palate in the United Kingdom

Journal:	*The Cleft Palate-Craniofacial Journal*
Manuscript ID	CPCJ-19-0132.R1
Manuscript Type:	Original Article
Keywords:	Feeding, Nursing, Parental perception, Psychological assessment, Psychosocial adjustment, Quality of life

Abstract:

Having a child born with a cleft lip and/or palate (CL/P) poses several challenges for new parents and can have a significant psychological impact on the family as a whole. Previous research has indicated that dissatisfaction with healthcare is a risk factor for poor parental adjustment and family functioning. Yet, knowledge is lacking in regard to which aspects of care parents may be dissatisfied with. The current study aimed to comprehensively evaluate healthcare satisfaction in families following a diagnosis of CL/P by utilising data collected from a UK-wide birth cohort.

Self-reported questionnaire data were obtained from 517 parent dyads enrolled in The Cleft Collective Birth Cohort Study. The 'Pediatric Quality of Life Inventory - Healthcare Satisfaction Generic Module' was used as the primary outcome measure.

Overall, parents were satisfied with the care they had received. However, less favourable scores were identified in relation to the information parents had been given. A good degree of agreement between mothers and fathers was observed. However, marginal evidence suggested that fathers were significantly more dissatisfied than mothers regarding the 'Communication' and 'Inclusion of Family' subscales.

Although the findings of this large-scale study reflect overall healthcare satisfaction, issues are raised in relation to the quality of information families received, particularly for fathers. In addition, fathers may feel less included in their child's treatment pathway. These findings offer practical suggestions as to which areas of care could be targeted by all health professionals to improve parents' healthcare experiences and promote overall familial adjustment.

Satisfaction with Healthcare in Families Following a Diagnosis of Cleft Lip and/or Palate in the United Kingdom

Abstract

Objective: Having a child born with a cleft lip and/or palate (CL/P) poses several challenges for new parents and can have a significant psychological impact on the family as a whole. Previous research has indicated that dissatisfaction with healthcare is a risk factor for poor parental adjustment and family functioning. Yet, knowledge is lacking in regard to which aspects of care parents may be dissatisfied with. The current study aimed to comprehensively evaluate healthcare satisfaction in families following a diagnosis of CL/P by utilising data collected from a UK-wide birth cohort.

Methods: Self-reported questionnaire data were obtained from 517 parent dyads enrolled in The Cleft Collective Birth Cohort Study. The 'Pediatric Quality of Life Inventory - Healthcare Satisfaction Generic Module' was used as the primary outcome measure.

Results: Overall, parents were satisfied with the care they had received. However, less favourable scores were identified in relation to the information parents had been given. A good degree of agreement between mothers and fathers was observed. However, marginal evidence suggested that fathers were significantly more dissatisfied than mothers regarding the 'Communication' and 'Inclusion of Family' subscales.

Conclusions: Although the findings of this large-scale study reflect overall healthcare satisfaction, issues are raised in relation to the quality of information families received, particularly for fathers. In addition, fathers may feel less included in their child's treatment pathway. These findings offer practical suggestions as to which areas of care could be

targeted by all health professionals to improve parents' healthcare experiences and promote overall familial adjustment.

Key words: cleft lip and palate; cohort study; healthcare satisfaction; treatment; The Cleft Collective; parents

Introduction

Background

From the point of diagnosis through to adulthood, care for individuals born with a cleft lip and/or palate (CL/P) is multidisciplinary and complex. In the UK, care is delivered via a standardised treatment pathway, designed to address both the functional and appearance-related consequences of CL/P (National Health Service, 2013). In the first year of life alone, infants may require several medical interventions, including respiratory and/or feeding support, surgery to repair the cleft, and the insertion of grommets.

Prior to the national centralisation of CL/P services in the early 2000s, UK cleft care was delivered in a largely uncoordinated manner, often leading to suboptimal patient outcomes (Sandy et al., 1998). Following the recommendations presented in the 1998 Clinical Standards Advisory Group report, care has since been refined to a smaller number of highly specialist sites. Further, and in recognition of the broader impact of CL/P, psychological support has been integrated as a core component of UK cleft care.

It is now widely recognised that CL/P can have a significant psychosocial impact on both the individual and the family as a whole. In a large-scale qualitative study exploring parents' experiences of caring for a child with CL/P, Nelson and colleagues (2012) identified several

emotional tensions that parents have to manage throughout the course of their child's treatment. The anticipation of surgical intervention was found to be particularly daunting, whereby parents sanction treatment to achieve optimal outcomes, yet also want to protect their children from discomfort and distress. Research has also highlighted the potential for treatment-related distress in parents; for example, intense discomfort from watching their child undergo general anaesthetic (Stock & Rumsey, 2015). The physical consequences of treatment, such as bleeding, swelling, and the dramatic change in their child's appearance following surgery can also have a great impact on parents (Nelson et al., 2012). Additionally, parents have discussed how treatment can be intensive, and disrupt family functioning (Stock & Rumsey, 2015). Taken together, previous findings point to the emotional strain associated with CL/P treatment, particularly during the first year of the child's life.

In a recent UK-wide quantitative study exploring parents' psychosocial adjustment following the birth of a child with CL/P (Stock et al., in press), satisfaction with healthcare was consistently associated with higher levels of parental psychological wellbeing. This relationship between healthcare satisfaction and parental adjustment reinforces the importance of high quality, coordinated, patient-centred care. Nonetheless, previous qualitative studies exploring the healthcare experiences of parents of children with CL/P highlight potential service-related issues (Knapke et al., 2010; Searle et al., 2016). To date, little research has explored parents' satisfaction with healthcare from a quantitative perspective (Stock & Feragen, 2016). This may reflect the understanding that healthcare satisfaction can be a challenging construct to measure, and that the collation of 'total scores' often results in a 'ceiling effect' (Moret et al., 2007). Although previous findings suggest that parents rate CL/P healthcare positively on the whole (Damiano et al., 2006; Nelson & Kirk, 2013; Feragen et al., 2017), the views of parents who are less satisfied remain unheard, and

knowledge of which particular aspects of care parents are satisfied and dissatisfied with is scarce.

Due to the complex nature of the UK CL/P treatment pathway and the potential for healthcare dissatisfaction to negatively impact parental wellbeing, a better understanding of the factors that contribute to a good healthcare experience is critical. The present study aimed to comprehensively evaluate healthcare satisfaction in families following a diagnosis of CL/P in their child, by utilising data collected from a UK-wide cohort study.

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 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 for 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 following the birth of their child, 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 transferred to the authors in an encrypted password-protected file.

Measures

The Pediatric Quality of Life Inventory: Healthcare Satisfaction Generic Module (PedsQL-HSGM; Varni et al., 2004) is a 24-item parent-reported measure assessing six dimensions of healthcare satisfaction (Information, 5 items; Inclusion of Family, 4 items; Communication, 5 items; Technical Skills, 3 items; Emotional Needs, 4 items; and Overall Satisfaction, 3 items). Items are rated on a 5-point Likert scale (0 = Never; 4 = Almost Always). Responses are reverse-scored as appropriate and linearly transformed to a 0-100 scale, whereby a higher score indicates greater satisfaction. In the current study, the Technical Skills and Emotional Needs subscales were not included, since the majority of items are not applicable in early infancy. For the same reason, only 3 items of the Communication subscale were included in the analyses. The Communication subscale includes questions relating to staff's communication and listening skills, and how well parents felt they were prepared for their child's treatment. The Information subscale includes questions about the quality and amount of information that the parents received. The Inclusion of Family subscale includes questions on staff sensitivity, staff willingness to discuss the family's concerns, and staff readiness to include the family in decision-making. The Overall Satisfaction subscale includes questions about how satisfied parents were with staff approachability, their child's treatment outcomes and the overall care that they and their child had received.

Additional bio-demographic data (such as child's cleft type) were primarily derived from parent-completed Cleft Collective questionnaires. Where parental consent had been given to

do so, key bio-demographic information was also extracted from the child's 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 analysis. There were no unduly large or strongly influential observations in the sample. A missing values analysis was performed. Any missing data would seem to be missing at random and the extent of this was not considered to unduly affect conclusions (Schafer, 1999). Since mothers and fathers were only moderately correlated across all subscales (Pearson correlation coefficients ranged from 0.45 to 0.49), the authors proceeded to conduct separate analyses for the two groups. First, PedsQL-HSGM subscale means were calculated and paired samples *t*-tests were performed to assess differences between mothers and fathers. A second series of paired-samples *t*-tests were conducted to test whether subscale means significantly differed from one another. For all *t*-tests, effect sizes were calculated in order to quantify any differences. For the paired samples *t*-tests, the Dunlap et al. (1996) method for calculating effect size was used. For tests of difference, *d* values between 0.1 and 0.2 indicate a small effect, values between 0.2 and 0.5 a moderate 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). Given the nature of the data, the Hodges-Lehman 95% confidence interval for the median difference was also calculated.

Results

Participants

Participants in this study comprised 517 parent dyads who contributed baseline questionnaire data to The Cleft Collective Birth Cohort Study between December 2013 and December 2017. 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 UK Cleft Registry and Audit Network in 2017.

Satisfaction with Healthcare

Table 2 shows mothers' and fathers' mean scores on the PedsQL-HSGM subscales. Results indicate that fathers were significantly less satisfied than mothers in terms of Information, Communication, and Inclusion of Family, with small effect sizes (d = 0.119, d = 0.097, d = 0.137 respectively). The 95% confidence intervals for the median differences indicate that differences between mothers' and fathers' scores is small. However, the percentage of participants reporting maximum satisfaction scores is particularly low in regard to the Information subscale (mothers = 39.9%, fathers = 35.3%), and is less than 65% on all domains. Scores are consistently lower for fathers than mothers on all four domains.

Mothers' and fathers' mean subscale scores were also compared to one another (Table 3). For both mothers and fathers, differences were statistically significant for the following pairs: Information and Communication; Information and Inclusion of Family; and Information and Overall Satisfaction. These findings indicate that both mothers and fathers scored significantly lower on the Information subscale when comparing to all other subscales. Effect

sizes were moderate overall (d = 0.301 to 0.343) and the 95% Confidence Interval for the median difference indicates small effects.

Discussion

This study utilised parent-reported data collected from a UK-wide birth cohort to comprehensively evaluate healthcare satisfaction in families following a diagnosis of CL/P. The findings of this study are discussed in more detail below, alongside recommendations for research and clinical practice.

Overall Satisfaction with Healthcare

On the whole, participants in the current study reported a high level of satisfaction with the healthcare that they and their child had received following a diagnosis of CL/P. This finding is in line with previous CL/P research (Damiano et al., 2006; Nelson & Kirk, 2013; Feragen et al., 2017) and the wider paediatric literature (Cousino & Hazen, 2013), and provides a positive overall view of the health services relevant to CL/P. Healthcare satisfaction is a key predictor of overall familial wellbeing (Stock et al., in press), and as such should be systematically monitored and maintained

Dissatisfaction with Patient Information

Despite reporting a high level of satisfaction overall, both mothers and fathers reported significantly less favourable scores on the Information subscale when compared to all other subscales. It is acknowledged that these differences are relatively small. However, maximum scores on the Information subscale occurred less than 40% of the time for both mothers and fathers, suggesting that the information families receive regarding their child's diagnosis and treatment pathway may not always address all of the questions they have. Similar findings

also arise in the general paediatric literature. For example, Miceli and Clark (2005) explored parental satisfaction with the paediatric inpatient experience and parents' suggestions for improvement and found that the improvement of healthcare information was a key priority for parents. Findings such as this emphasise the importance of providing personalised information to parents wherever possible, in which content, format, and timing is based on individual preferences. It is imperative that the appropriate level of information is achieved, since negative interactions can have a profound impact on parents during a time of emotional turmoil and uncertainty and can impede longer-term familial adjustment (Cousino & Hazen, 2013). Previous CL/P literature has identified the potential psychological impact of a poorly handled diagnosis and/or suboptimal postnatal care as delivered by non-specialist health professionals, such as sonographers and midwives (Knapke et al., 2010; Searle et al., 2016; Stock et al., 2019). Within this literature, a lack of knowledge, information and adequate support is a reoccurring theme. Comprehensive suggestions for improvements, including increased training and support for non-specialist health professionals, widespread access to reliable information in a variety of formats, and a closer collaboration between specialist CL/P teams and non-specialists have been previously offered (Searle et al., 2016; Stock et al., 2019).

Inclusion of Fathers in Healthcare

Despite relatively small effects, fathers reported significantly less favourable scores in relation to the Communication, Inclusion of Family, and Information subscales when compared to mothers. Further, fathers reported significantly lower scores on the Information subscale in comparison to all other subscales. Previous qualitative studies have shown that fathers often report their information needs are not being satisfactorily addressed, and that they can feel excluded from maternity services (Stock & Rumsey, 2015; Zeytinoglu et al., 2016). Wider literature has demonstrated that fathers tend to adopt a supportive and

information-seeking role within the family (Ahmann, 2006; Deeney et al., 2009), and may therefore feel particularly distressed if they do not feel they are receiving adequate information regarding their child's health. In addition to healthcare satisfaction, positive communication is associated with adherence to treatment protocols and clinic visits (Zolnierek & diMatteo, 2009). It is therefore imperative that fathers are fully included in their family's treatment journey, by ensuring equal access to information, giving fathers space to communicate any questions or concerns, taking these concerns seriously and acting upon them, and communicating any treatment plans clearly and consistently, to avoid fathers slipping through the net in the longer-term. Fathers not only require access to information and support for themselves, but also play a key role in the upbringing of their child. The findings of the present study therefore lend further support to the importance of routinely assessing paternal as well as maternal wellbeing, and utilising family-centred approaches in both clinical care and research (Stock & Rumsey, 2015; Zeytinoglu et al., 2016).

Methodological Considerations

In a review of the literature, Nelson and colleagues (2012) note that previous studies in the CL/P field have been limited by data collected from small samples and/or single sites. In addition, most of the existing work on healthcare satisfaction in CL/P has tended to utilise qualitative methodology, preventing more generalisable conclusions. To the authors' knowledge, this is the largest quantitative study worldwide to date to examine parental healthcare satisfaction in the period following a diagnosis of CL/P. A further strength of the current sample is a high representation of fathers, who have historically been unrepresented in CL/P research (Nelson et al., 2012; Stock & Rumsey, 2015).

The current study focused on healthcare satisfaction in parent dyads. However, single parent families are also of interest to the field. Future CL/P research could aim to be inclusive of all

family types, and to examine any differences in outcomes to assess whether single parent families may represent a group with elevated psychosocial risk, as has been found in the broader literature.

Data were collected using a validated and widely used measure of healthcare satisfaction (Varni et al., 2004). One of the benefits of this measure is that it consists of several dimensions. Scores are therefore indicative of areas of particular satisfaction/dissatisfaction, in addition to the overall satisfaction score. In the current study, overall satisfaction was found to be high, yet areas of concern were identified when exploring the data in more depth. Although not all items of the measure could be utilised in the current study due to the young age of the participants' children, the PedsQL-HSGM appears to be a useful measure in directing health professionals to areas which may be in need of improvement, as well as areas of strength.

Data were extracted from The Cleft Collective Birth Cohort study (Stock et al., 2015). 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 household income. A lack of a more culturally diverse sample is not uncommon in health literature; however, it means that some individuals' experiences of healthcare will have been missed. Literature from a range of health fields has demonstrated clear differences in the way some minority groups interact with health services and engage with research (Public Health England, 2017). Further work is therefore needed in order to better understand the needs and healthcare experiences of these potentially vulnerable groups in relation to CL/P, and to ensure that studies are relevant and accessible.

Another methodological consideration is that participants were asked to report upon their healthcare experiences up until the point of primary surgery. This is a relatively large timescale, and for many parents could encompass both antenatal and postnatal care. The questions asked also do not differentiate between the care delivered by CL/P specialists and by non-specialists; rather they ask parents to reflect on their overall journey to date. In qualitative studies, parents of children with CL/P have identified some differences, often praising CL/P specialists and criticising non-specialists (Jeffery & Boorman, 2001; Knapke et al., 2010; Lindberg & Berglund, 2014; Searle et al., 2016). Nonetheless, affected families are the responsibility of the health service as a whole, and patient-centred healthcare involves the establishment of partnerships between all providers.

Last, the data explored in the current study were cross-sectional in nature. This limits the conclusions that can be drawn, particularly given that treatment for CL/P will run into adulthood, and that healthcare satisfaction is liable to fluctuate over time (Nelson & Kirk, 2013). The impact of healthcare satisfaction on longer-term outcomes is also of interest to the field. As research programmes such as The Cleft Collective develop, longitudinal analyses of this kind will become possible.

Conclusions

The present study explored satisfaction with healthcare in families following a diagnosis of CL/P in their child. Findings align with previous CL/P research and suggest that overall, parents are satisfied with the healthcare they receive. However, findings also highlight some parental dissatisfaction regarding the information received regarding their child's condition, particularly for fathers. Fathers may also feel less included in the healthcare process. The findings of this large-scale study offer practical suggestions as to which areas of care could

be targeted by healthcare professionals to improve parents' experiences and promote overall familial psychological adjustment.

References

Ahmann E. Supporting fathers' involvement in children's health care. *Pediatr Nurs.* 2006;32:88-90.

Byrnes AL, Berk NW, Cooper ME, Marazita ML. Parental evaluation of informing interviews for cleft lip and/or palate. *Pediatrics.* 2003;112(2):308.

Cleft Registry & Audit Network (2017). *Annual report on cleft lip and/or palate 2017*. Royal College of Surgeons, United Kingdom.

Cohen J. Statistical Power Analysis for the Behavioural Sciences (2nd edition). Hillsdale, NJ: Erlbaum. 1988.

Cousino MK, Hazen RA. Parenting stress among caregivers of children born with chronic illness: a systematic review. *J Pediatr Psychol*. 2013;38(8):809-828.

Damiano PC, Tyler MC, Romitti PA, Momany ET, Canady JW (…), Murray JC. Type of oral cleft and mothers' perceptions of care, health status, and outcomes for preadolescent children. *Cleft Palate Craniofac J.* 2006;43:715-721.

Deeney K, Lohan M, Perkes J, Spence D. Experiences of fathers of babies in intensive care. *Pediatr Nurs.* 2009;21:45-47.

Dunlap WP, Cortina JM, Vaslow JB, Burke MJ. Meta-analysis of experiments with matched groups or repeated measures designs. *Psychol Methods*. 1996;1(2):170-177.

Feragen KB, Semb G, Heliövaara A, Lohmander A, Johannessen EC (…), Rumsey N. Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: 10. Parental perceptions of appearance and treatment outcomes in their 5-year-old child. *J Plast Surg Hand Surg*. 2017;51:81-87.

Johansson B, Ringsberg KC. Parents' experiences of having a child with cleft lip and palate. *J Adv Nurs*. 2004;47(2):165-173.

Lindberg N, Berglund AL. Mothers' experiences of feeding babies born with cleft lip and palate. *Scand J Caring Sci.* 2014;28:66-73.

Knapke SC, Bender P, Prows C, Schultz JR, Saal HM. Parental perspectives of children born with cleft lip and/or palate: a qualitative assessment of suggestions for healthcare improvements and interventions. *Cleft Palate Craniofac J.* 2010;47(2):143-150.

Miceli PJ, Clark PA. Your patient – my child: seven priorities for improving pediatric care from the parents' perspective. *J Nurs Care Qual*. 2005;20:43-53.

Moret L, Nguyen JM, Pillet N, Falissard B, Lobrail P, Gasquet I. Improvement of psychometric properties of a scale measuring inpatient satisfaction with care: a better response rate and a reduction of the ceiling effect. *BMC Health Serv Res*. 2007;7:197.

National Health Service. NHS standard contract for cleft lip and/or palate services including non-cleft velopharyngeal dysfunction (VPD) (all ages). 2013. https://www.england.nhs.uk/wp-content/uploads/2013/06/d07-cleft-lip.pdf Accessed 16 October, 2018.

Nelson P, Glenny AM, Kirk, S, Caress AL. Parents' experiences of caring for a child with a cleft lip and/or palate: a review of the literature. *Child Care Health Dev.* 2012;38:6-20.

Nelson PA, Kirk SA, Caress AL, Glenny AM. Parents' emotional and social experiences of caring for a child through cleft treatment. *Qual Health Res.* 2012;22(3):346-359.

Office for National Statistics. Statistical bulletin: household disposable income and inequality in the UK: financial year ending 2017. 2017. https://www.ons.gov.uk/peoplepopulationandcommunity/personalandhouseholdfinances/incomeandwealth/bulletins/householddisposableincomeandinequality/financialyearending2017 Accessed 16 October, 2018.

Public Health England. Health Profile for England – chapter 6: Social determinants of health. 2017. www.gov.uk/government/publications/health-profile-for-england/chapter-6-social-determinants-of-health Accessed 16 October, 2018.

Sandy JR, Williams AC, Mildinhall S, Murphy T, Bearn D, Shaw WC, Sell D, Devlin M, Murray JJ. The Clinical Standards Advisory Group (CSAG) cleft lip and palate study. *Br J Orthod*. 1998;25:21-30.

Schafer JL. Multiple imputation: A primer. *Stat Methods Med Res*. 1999;8:3–15.

Searle A, Ryan S, Waylan A. Health professional communication and the diagnosis and care of infants born with cleft lip and palate in the UK. *J Neonatal Nurs*. 2016;22(5):236-243.

Stock NM, Costa B, Williams JR, Martindale A, VTCT Foundation Research Team at the Centre for Appearance Research. Breaking the news: Parents' experiences of receiving an antenatal diagnosis of cleft lip. *Cleft Palate Craniofac J.* 2019; doi: 10.1177/1055665619830884.

Stock NM, Costa B, White P, Rumsey N. Risk and protective factors for psychological distress in families following a diagnosis of cleft lip and/or palate. *Cleft Palate Craniofac J*. In press.

Stock NM, Feragen, KB. Psychological adjustment to cleft lip and/or palate: A narrative review of the literature. *Psychol Health*. 2016;31(7):777-813.

Stock NM, Humphries K, Pourcain BS, Bailey M, Persson M, Ho KM, Ring S, Marsh C, Albery L, Rumsey N, Sandy J. Opportunities and Challenges in Establishing a Cohort Study: An Example From Cleft Lip/Palate Research in the United Kingdom. *Cleft Palate Craniofac J*. 2016;53(3):317-325.

Stock NM, Rumsey N. Parenting a child with a cleft: the fathers' perspective. *Cleft Palate Craniofac J.* 2015;52(1):31-43.

Varni JW, Burwinkle TM, Dickinson P, Sherman SA, Dixon P (…), Sadler BL. Evaluation of the built environment at a children's convalescent hospital: Development of the Pediatric Quality of Life Inventory parent and staff satisfaction measures for pediatric health care facilities. J Dev Behav Ped. 2004;25:10-20.

Young JL, O'Riordan M, Goldstein JA, Robin, NH. What information do parents of new borns with cleft lip, palate, or both want to know? *Cleft Palate Craniofac J.* 2001;38:55-58.

Zeytinoglu S, Davey MP, Cerand C, Fisher K. Fathers of children born with cleft lip and palate: impact of timing of diagnosis. *Fam Syst Health.* 2016;34(2):150-158.

Zolnierek KB, di Matteo MR. Physician communication and patient adherence to treatment: A meta-analysis. *Med Care*. 2009;47:826-834

Table 1: Participant Characteristics			
Parent Characteristics			**UK Census Data**
	Mothers	**Fathers**	
Mean (SD) Age at Conception	30.83 (5.462)	33.56 (7.318)	
Annual Gross Income			£28,677
0-£19,999	59.1%	27.3%	
£20,000-£39,999	34.6%	49.0%	
£40,000-£59,999	4.1%	12.1%	
£60,000+	2.3%	11.7%	
Education			
No qualifications	2.0%	3.6%	23.0%
School-level qualifications	37.4%	44.4%	44.0%
Undergraduate degree or above	50.5%	34.8%	27.0%
Other	10.1%	13.7%	6.0%
Country of Birth			
UK	79.4%	82.8%	86.0%
Other	20.6%	17.2%	14.0%
Ethnicity			
White	90.6%	91.1%	86.0%
Other	9.4%	8.9%	14.0%
Child Characteristics			**CRANE Data**

Cleft Type		
Cleft Palate	34.3%	39.4%
Cleft Lip	24.0%	21.6%
Cleft Lip + Palate	41.8%	27.6%
Unregistered		11.4%

Table 2: Mothers' and Fathers' Mean Subscale Scores and 95% Confidence Interval for Median of the Differences between Mother and Father

	Mother		Father		Comparison	
PedsQL-HSGM Subscale	**Mean (SD)**	**Percentage Scoring 100% (95% CI)**	**Mean (SD)**	**Percentage Scoring 100% (95% CI)**	**t-test**	**95% CI for Med Diff**
Information	84.6 (18.9)	40.7 (36.5 to 45.1)	83.2 (19.5)	35.3 (31.1 to 39.6)	$t(512)=2.634$, $p=0.009$** $d = 0.119$	0.0 to 2.5
Communication	90.0 (17.2)	64.6 (60.1 to 69.0)	89.1 (16.6)	58.5 (53.9 to 63.1)	$t(457)=2.052$, $p=0.041$* $d = 0.097$	0.0 to 0.0
Inclusion of Family	90.4 (16.1)	61.0 (56.5 to 65.3)	88.9 (16.8)	56.4 (52.0 to 60.8)	$t(504)=2.934$, $p=0.003$** $d = 0.137$	0.0 to 0.0
Overall Satisfaction	89.7 (16.1)	61.2 (56.5 to 65.7)	88.9 (15.8)	58.3 (53.7 to 62.9)	$t(455)=1.589$, $p=0.113$ $d = 0.078$	0.0 to 0.0

*** $p < 0.001$, ** $p < 0.01$, * $p < 0.05$

Table 3: Comparison of Subscale Means

Pairs	Mothers *t*-tests	Fathers *t*-tests
Information - Communication	*t*(638)=-10.261, *p*<0.001*** *d* = 0.320 *Md = -4.2 [-5.0 to -3.3]*	*t*(506)=-10.385, *p*<0.001*** *d* = 0.321 *Md = -4.2 [-5.0 to -3.1]*
Information - Inclusion of Family	*t*(675)=-11.336, *p*<0.001*** *d* = 0.343 *Md = -4.2 [-5.0 to -3.1]*	*t*(528)=-9.846, *p*<0.001*** *d* = 0.310 *Md = -3.8 [-5.0 to -2.5]*
Communication - Inclusion of Family	*t*(642)=-0.803, *p*=0.422 *d* = 0.018 *Md = 0.0 [0.0 to 0.0]*	*t*(516)=-0.082, *p*=0.935 *d* = 0.002 *Md = 0.0 [0.0 to 0.0]*
Communication – Overall Satisfaction	*t*(598)=0.520, *p*=0.603 *d* = 0.016 *Md = 0.0 [0.0 to 0.0]*	*t*(488)=0.243, *p*=0.808 *d* = 0.008 *Md = 0.0 [0.0 to 0.0]*
Information – Overall Satisfaction	*t*(634)=-7.645, *p*<0.001*** *d* = 0.301 *Md = -4.2 [-5.0 to -2.5]*	*t*(496)=-7.804, *p*<0.001*** *d* = 0.317 *Md = -5.0 [-5.83 to -2.5]*
Inclusion of Family – Overall Satisfaction	*t*(631)=1.568, *p*=0.117 *d* = 0.051 *Md = 0.00 [0.0 to 0.0]*	*t*(505)=-0.084, *p*=0.933 *d* = 0.003 *Md = 0.0 [0.0 to 0.0]*

***<0.001, **<0.01, *<0.05

p-values given are unadjusted for multiplicity of tests. Results with p < 0.05 remain statistically significant after a Bonferonni correction.

Full list of research variables	
Information about parents	Ethnicity Marital status Age at conception Annual income Living in UK since birth Highest educational qualification
Information about child	Cleft type Timing of cleft diagnosis
Outcome measure - Peds-QL HSGM	Technical subscale Information subscale Communication subscale (3 items only) Inclusion subscale Emotional subscale Overall subscale
A data dictionary detailing the variables collected in The Cleft Collective Birth Cohort Study is available at: www.bristol.ac.uk/cleft-collective/professionals/access.	