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Parent-reported socioemotional and cognitive development in children with a cleft lip and/or palate at 18 months: Findings from a UK birth cohort

Bruna Costa[1] | Paul White[2] | James D. Kiff[1] | Amy Davies[3] | Nicola M. Stock[1]

[1]Centre for Appearance Research, University of the West of England, Bristol, UK

[2]Applied Statistics Group, University of the West of England, Bristol, UK

[3]The Cleft Collective, The University of Bristol, Bristol, UK

Correspondence
Bruna Costa, Centre for Appearance Research, University of the West of England, Coldharbour Lane, Bristol BS16 1QY, UK.
Email: bruna.oliveiracosta@uwe.ac.uk

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Abstract

Background: One of the most common congenital conditions in the world, cleft lip and/or palate (CL/P) has been shown to potentially impact long-term physical and developmental outcomes in affected children. However, little is known about the factors that contribute to such outcomes, and there is a lack of consensus about which screening tools may be most effective. The aims of the current study were (a) to assess parent-reported socioemotional and cognitive development in children born with CL/P at 18 months of age; (b) to identify factors associated with the incidence of developmental concerns; and (c) to assess the utility of the widely recommended Ages and Stages Questionnaires (ASQs) in identifying developmental concerns from an early age in the CL/P population.

Methods: Parent-reported questionnaire data were extracted from The Cleft Collective Cohort Study for 322 mothers of children with CL/P aged 18 months.

Results: Mean scores across both ASQ measures indicated typical development in the study sample overall. However, 31.1% of children met a referral criterion on at least one domain. Child-related risk factors included problems with physical development and feeding method. Parent-related risk factors included the mother's levels of anxiety and depression and mother's marital status. Additional developmental concerns extracted from mothers' qualitative data included feeding difficulties, speech development, sleep patterns, aggressive behaviours, vision, oral health, hearing, breathing and motor skills.

Conclusions: The majority of children in this study were developing as expected at 18 months of age. However, parent-reported developmental concerns were identified in a minority of children, suggesting a need to screen for potential risk factors in routine practice. Further, the ASQ appears to offer a viable option in the early identification of developmental concerns in children with CL/P. A combined medical and systemic approach to healthcare is recommended to support the prevention of long-term developmental concerns in the child and poor psychological adjustment in parents.

KEYWORDS

Ages and Stages Questionnaire, child development, cleft lip and palate, parental well-being, risk and protective factors, The Cleft Collective

1 | INTRODUCTION

One of the most common congenital conditions in the world, cleft lip and/or palate (CL/P) affects 1 in every 1,000 live births (World Health Organization, 2018). CL/P may be caused by a genetic predisposition, as well as environmental factors such as maternal health behaviours and practices during pregnancy (Mossey, Little, Munger, Dixon, & Shaw, 2009). In some cases, CL/P can be associated with additional conditions and syndromes such as Pierre Robin sequence and DiGeorge syndrome (Dixon, Marazita, Beaty, & Murray, 2011). In most high-income countries, the cleft is typically surgically repaired during the first year of the infant's life, but those affected may also require subsequent surgeries throughout childhood and into adulthood (including elective and revision surgeries). Families of children with CL/P are therefore expected to engage with a long-term multidisciplinary treatment pathway to address the functional and appearance-related consequences of the condition. This may include psychological support, speech and language therapy, and orthodontics, among other treatments (National Health Service [NHS] England, 2013). As with most chronic health conditions, CL/P can impact the child's emotional, social and cognitive development (Stock & Feragen, 2016), as well as the wider family's well-being and functioning (Nelson, Glenny, Kirk, & Caress, 2012).

Previous research has indicated that children with CL/P are in poorer overall health than the general population (Stock & Feragen, 2016), with an increased risk of special health care needs (Damiano et al., 2006), hospitalization (Wehby, Pedersen, Murray, & Christensen, 2012) and mortality (Christensen, Juel, Herskind, & Murray, 2004). Studies have also identified a higher percentage of developmental and behavioural concerns as well as poorer academic performance among children with CL/P compared with their unaffected peers (Burnell et al., 2014; Chetpakdeechit et al., 2011; Feragen & Stock, 2014; Hunt, Burden, Hepper, Stevenson, & Johnston, 2007; Knight, Cassell, Meyer, & Strauss, 2015; Murray et al., 2010; Richman, McCoy, Conrad, & Nopoulos, 2012; Tillman et al., 2018; Wehby et al., 2014). Yet the mechanisms that account for such developmental differences remain largely unknown, particularly in younger children with whom research is sparse (Murray et al., 2010). It is possible that neuropsychological patterns associated with CL/P impact development; however, knowledge of this is limited in the very early years of life (Richman et al., 2012). It is also possible that rather than the cleft itself, early parent–child interactions and familial characteristics are key influences for later developmental concerns (Hunt et al., 2007; Murray et al., 2010). Hunt and colleagues argue that if a child has a visible facial scar as a result of the cleft, this may adversely influence the ways in which the parents perceive and interact with him/her. Further, even very young children with CL/P, particularly when the palate is involved, can have poorer communication skills (including hearing loss) and problems related to speech intelligibility (Broen, Devers, Doyle, Prouty, & Moller, 1998; Schönweiler et al., 1999), which may limit communication and further influence the parent–child dynamic. Worryingly, poor familial relationships and attachment have been found to predict a range of difficulties in children and increase their vulnerability to adverse outcomes (Despars et al., 2011).

Key messages

- Children born with cleft lip and/or palate may be at risk of parent-reported developmental concerns if additional physical anomalies are present, if fed using a nasogastric tube alongside formula milk, if raised in a single-parent family and if the mother's levels of anxiety and/or depression are elevated.
- The use of the Ages and Stages Questionnaires could be considered in routine clinical practice and in craniofacial research to identify at-risk families from an early age.
- To prevent long-term developmental concerns in the child and poor psychological adjustment in parents, a combined medical and systemic approach to healthcare is advocated.

Taken together, the literature implies that early screening for developmental concerns in children born with CL/P may be warranted in order to provide appropriate support and prevent long-term disadvantage. However, inconsistencies in efforts to identify developmental concerns have been indicated. For example, some but not all CL/P teams in the United Kingdom reported using formal developmental assessments, and only a minority of teams reported screening for developmental concerns routinely (Stock, MacLeod, & Clayton-Smith, 2019). Equally, several different outcome measures have been proposed for use in the craniofacial population (Collett & Speltz, 2006), and there is a lack of consensus over which tools are most useful and when (Stock et al., 2019). The Ages and Stages Questionnaires (ASQs) are widely used in early educational and healthcare settings more broadly and are endorsed by the UK government (Ofsted & NHS Digital, 2017). These brief, parent-reported measures may therefore also be useful in early identification of developmental concerns in CL/P.

The aims of the present study were to (a) assess parent-reported socioemotional and cognitive development in children born with CL/P at 18 months of age in comparison with the general population; (b) identify potential contributory factors for such developmental concerns; and (c) assess the utility of the ASQ in identifying early developmental concerns in the CL/P population.

2 | METHODS

2.1 | Procedure

Ethical approval to establish The Cleft Collective Cohort Studies was granted by the South West Central Bristol Ethics Committee (REC approval 13/SW/0064). Global research and development (R&D) approval was provided by the University Hospitals Bristol. Local R&D

approvals were subsequently obtained from each 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. Handwritten informed consent was then obtained from the parents. Parents were specifically asked for permission to use their provided data in the future for individual ethically approved research studies.

Participants completed The Cleft Collective 18-month questionnaire when their child was between 17 and 19 months of age 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 the University of the West of England. Confidentiality agreements to access the data were signed by the authors, and data were subsequently de-identified and transferred to the authors in an encrypted password-protected file.

2.2 | Outcome measures

The ASQs (Ages and Stages Questionnaire-3 [ASQ-3; Squires & Bricker, 2009] and Ages and Stages Questionnaire: Social–Emotional [ASQ-SE; Squires, Bricker, & Twombly, 2002]) were used to screen for potential socioemotional and cognitive developmental concerns in children at 18 months of age, as reported by mothers.

The ASQ-3 is a 37-item measure used to assess child development in five core domains: communication skills (e.g., 'Can your child say eight or more words in addition to "mama" and "dada"?'); gross motor skills (e.g., 'Does your child walk well and seldom fall?'); fine motor skills (e.g., 'Can your child stack a small block or toy on top of another one?'); problem-solving (e.g., 'Does your child scribble back and forth when you give him/her a crayon?'); and personal–social skills (e.g., 'Does your child copy the activities you do?'). Parents are asked to score each item as 'yes', 'sometimes' or 'not yet'. ASQ-3 guidelines state that children who score at or below the cut-off may warrant a referral for further assessment, and children who score within the monitoring zone may require additional observation and a subsequent review of progress.

The ASQ-SE is a 29-item measure used to assess children's socioemotional development (e.g., 'Does your child look at you when you talk to him/her?'). ASQ-SE guidelines state that children who score on or above the cut-off may warrant a referral for further assessment.

2.3 | Additional variables of interest

Demographic information alongside additional variables that could potentially be considered to contribute to child development at 18 months were extracted from The Cleft Collective 18-month questionnaires.

Data collected from standardized measures were also examined. Most pertinent to the current study is the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983). The HADS is a 14-item self-reported measure of common 'symptoms' related to anxiety and depression experienced during the last month. Participants are asked to respond to each item using a 4-point Likert scale (e.g., 0 = *not at all*; 3 = *most of the time*), where higher scores indicate a higher level of emotional distress. A score of 0–7 is considered 'normal', whereas a score of 8–10 is considered 'borderline', and a score of 11 + is considered to be of 'clinical concern'.

A data dictionary detailing the variables collected in The Cleft Collective is available at www.bristol.ac.uk/cleft-collective/professionals/access.

2.4 | Analysis

For each of the five core domains, ASQ-3 outcome data are based on an ordinal 3-point scale ('normal', 'monitor' and 'referral'), and the ASQ-SE is a dichotomous outcome ('normal' and 'referral'). A combined measure of the number of times meeting referral thresholds and meeting at least once referral may be derived. The potential correlates of ASQ-3 and ASQ-SE under consideration are also categorical; they are predominantly nominal data (e.g., gender) or ordered categorical data (e.g., mother's anxiety determined through HADS labelled as 'normal', 'borderline' and 'clinical concern'). For these reasons, the primary analytical tool was a cross-tabulation with association examined using Pearson's chi-square test, with trend examined using the linear-by-linear first-order component (one degree of freedom) and with the strength of association quantified using Cramer's *V*. An r by c contingency table comprises r rows and c columns, and the degrees of freedom for the chi-square statistics are $(r-1)(c-1)$. Statistical thresholds for interpreting the magnitude of Cramer's *V* vary according to the degrees of freedom for the chi-square statistic. Cohen (1988) proffered lower bound thresholds of $V = .1$ (small), $V = .30$ (medium) and $V = .50$ (large) for one degree of freedom; $V = .07$ (small), $V = .21$ (medium) and $V = .35$ (large) for two degrees of freedom; $V = .06$ (small), $V = .17$ (medium) and $V = .29$ (large) for three degrees of freedom; and $V = .05$ (small), $V = .15$ (medium) and $V = .25$ (large) for four degrees of freedom (see Cohen, 1988). Structurally, Cramer's *V* is related to the chi-square statistic, χ^2, by $V = \sqrt{(\chi^2/nq)}$, where n is the sample size and $q = \min(r-1, c-1)$. Cramer's *V* for association and for linear-by-linear trend is summarized in the results along with the p value for association and trend. For 2×2 cross-tabulations, the strength of association was further quantified using the Cochran–Mantel–Haenszel (CMH) odds ratio and its 95% confidence interval (CI).

Cross-tabulations were used to screen factors potentially associated with developmental concerns. For a valid chi-square analysis, the expected cell value should be 5 or more in at least 80% of the cells, and no cell should have an expected value of <1. A factor was retained in the analysis if (a) it was associated ($p < .1$) with at least one outcome measure, and (b) it did not have more than 10%

missing data. Factors meeting the inclusion criteria are presented in Section 3, showing their relationship with at least one referral outcome.

Binary logistic regression was used to undertake a multivariate assessment of child specific factors (early health issues, problems with physical development, problems with psychological development, cleft type, gender, need for nasogastric feeding tube [NGT] assistance and feeding regime) on meeting at least one referral criterion.

3 | RESULTS

3.1 | Participants

A total of 322 mothers of children born with CL/P contributed 18-month questionnaire data to The Cleft Collective Birth Cohort Study between January 2015 and September 2018. In line with UK census data (Office for National Statistics, 2018), participating mothers were found to be predominantly White (94.7%) and UK born (84.0%). Of the current sample, 45.1% reported being educated to an undergraduate level or above, compared with 27.0% of the general population. The majority of mothers were married, in a civil union or in a domestic partnership (94.1%), which is higher than figures reported in the UK census (51.1%). Mothers in the current sample had a mean age of 32.7 years (*SD* = 5.4 years) at the time of data collection.

The mean gestational age of participating children was 39.7 weeks (*SD* = 2.2 weeks). In accordance with ASQ guidelines, data on children who were born prematurely (<37 weeks) were excluded. Children's mean age at the time of questionnaire completion was 18.3 months (*SD* = 0.5 months). A total of 57.8% of the children were male. Children's cleft type included cleft lip only = 21.3%; cleft palate only (including submucous cleft palate) = 37.8%; and cleft lip and palate = 40.9%. This distribution was found to be comparable with the national data reported by the Cleft Registry and Audit Network (CRANE) in 2017.

3.2 | Referral and monitoring zones

Table 1 shows children's mean scores on the ASQ-3and ASQ-SE, along with given cut-offs and monitoring zones for comparison where available. Mean scores for all five ASQ-3 domains indicated typical development in the study sample overall. The sample also displayed typical socioemotional development overall according to the mean scores obtained using the ASQ-SE.

Table 2 presents the percentages of children in the current study who scored within the cut-off for referral (ASQ-3 and ASQ-SE) and within the monitoring zone (ASQ-3 only). In total, 68.9% of children (222/322) did not meet referral criteria on any domain. A total of 17.7% children (57/322) met referral criteria on one domain, 7.8% (25/322) met referral criteria on two domains, and 3.4% (11/322) met referral criteria on three domains. None of the children met referral criteria on four domains, whereas 2.2% (7/322) met the referral criteria on five or more domains.

3.3 | Factors associated with children's development

Table 3 summarizes the relationship between potential associates for meeting at least one referral criterion. Child-related risk factors

TABLE 2 Study children in cut-off/monitoring zones

	Cut-off for referral	Monitoring zone
ASQ-3		
Communication	9.3% (*n* = 30)	46.3% (*n* = 149)
Gross motor skills	11.5% (*n* = 37)	10.9% (*n* = 35)
Fine motor skills	5.6% (*n* = 18)	5.3% (*n* = 17)
Problem-solving	15.5% (*n* = 50)	14.0% (*n* = 45)
Personal–social	7.1% (*n* = 23)	10.6% (*n* = 34)
ASQ-SE	5.6% (*n* = 18)	N/A

Abbreviations: ASQ-3, Ages and Stages Questionnaire-3; ASQ-SE, Ages and Stages Questionnaire: Social–Emotional; N/A, not applicable.

TABLE 1 ASQ-3 and ASQ-SE mean scores

	Current sample means (*SD*)	*n*	ASQ standard for referral	ASQ-3 standard for monitoring
ASQ-3[a]				
Communication	31.39 (14.479)	313	13.06	13.06–30.00
Gross motor skills	51.43 (13.998)	312	37.38	37.38–46.42
Fine motor skills	53.15 (10.595)	305	34.32	34.32–43.38
Problem-solving	40.69 (14.426)	275	25.74	25.74–35.86
Personal–social	44.92 (10.729)	300	27.19	27.19–37.55
ASQ-SE[b]	23.89 (17.117)	217	50	—

Abbreviations: ASQ-3, Ages and Stages Questionnaire-3; ASQ-SE, Ages and Stages Questionnaire: Social–Emotional; SD, standard deviation.
[a]Higher scores on the ASQ-3 indicate that a child's development appears to be on schedule.
[b]Lower scores on the ASQ-SE indicate that a child's development appears to be on schedule.

included problems with physical development, $\chi^2(1) = 10.26$, $p = .001$, and feeding method, the type of milk given to the child and/or the need for an NGT; $\chi^2(3) = 18.35$, $p < .001$.

Parent-related risk factors included the mother's level of anxiety, $\chi^2(2) = 9.33$, $p = .009$; the mother's level of depression, $\chi^2(2) = 15.06$, $p = .001$; and the mother's marital status, $\chi^2(1) = 5.00$, $p = .025$.

No clear effects of gender or cleft type were identified. Children who were identified by their parents as having problems with physical development were 2.9 times more likely to meet at least one referral criterion (CMH = 2.85, 95% CI 1.473–5.509). There was a marked increase in the referral rate for children who received formula milk and needed additional NGT support (48.8%) compared with breast milk only (22.6%), formula milk only (27.3%) or those who received breast milk and were in need of additional NGT support (6.7%).

The same statistical conclusions are obtained in a multivariable assessment using binary logistic regression. Specifically, in a logistic model, gender ($p = .382$), type of cleft ($p = .254$), previous health issues ($p = .993$) and problems with physical development ($p = .105$) showed no evidence to suggest a relation with meeting at least one referral criterion. However, after controlling for these factors, children with a parent-reported developmental concern were estimated to be 6.1 times more likely to meet at least one referral threshold ($p = .010$, 95% CI 1.53–24.17) than those without a parent-reported developmental concern. Children who required NGT assistance and were bottle-fed were just as likely to meet a referral criterion as children who were solely breastfed ($p = .172$). Children who required NGT assistance and were bottle-fed were 2.1 times more likely to meet at least one referral criterion than children who required NGT assistance and were breastfed ($p = .024$, 95% CI 1.37–90.1). They were also 2.0 times more likely to meet at least one referral criterion than children who did not required NGT and were otherwise bottle-fed ($p = .022$, 95% CI 1.11–3.70).

3.4 | Mother's concerns for their child's development

Mothers were also invited to provide additional comments about any concerns they had regarding their child's development in a free-text box. These qualitative comments were analysed by the first and fifth

TABLE 3 Associations meeting at least one threshold criterion

		No referral	At least one referral	χ^2, Cramer's *V* and CMH odds ratio
Child-related risk factors				
Child has problems with physical development[a]	Yes	47.6% (20)	52.4% (22)	$\chi^2(1) = 10.259$, $V = .176$, CMH = 2.849 (1.473–5.509), $p = .001$
	No	72.1% (202)	27.9% (78)	
Child's cleft type and gender	Lip male	80.5% (33)	19.5% (8)	$\chi^2(5) = 11.012$, $V = .184$, $p = .051$
	Lip female	80.0% (20)	20.0% (5)	
	Palate male	52.3% (23)	47.7% (21)	
	Palate female	63.5% (47)	36.5% (27)	
	Lip and palate male	71.0% (71)	29.0% (29)	
	Lip and palate female	72.4% (21)	27.6% (8)	
Type of milk given to child and need for NGT[b]	Breast only	77.4%	22.6%	$\chi^2(3) = 18.349$, $V = .235$, $p = .000$
	Formula or combination	72.7%	27.3%	
	Breast + NGT	93.3%	6.7%	
	Formula or combination + NGT	51.2%	48.8%	
Parent-related risk factors				
Mother's marital status	Single	50.0% (13)	50.0% (13)	$\chi^2(1) = 5.002$, $V = .126$, CMH = 0.406 (0.18–0.913), $p = .025$
	Partnered	71.1% (202)	28.9% (82)	
Mother's self-reported level of anxiety—from HADS	Normal	72.4% (152)	27.6% (58)	$\chi^2(2) = 9.328$, $V = .173$, $p = .009$
	Borderline	67.9% (36)	32.1% (17)	
	Clinical concern	47.4% (180)	52.6% (20)	
Mother's self-reported level of depression—from HADS	Normal	73.0% (176)	27.0% (65)	$\chi^2(2) = 15.056$, $V = .217$, $p = .001$
	Borderline	45.8% (22)	54.2% (26)	
	Clinical concern	53.3% (8)	46.7% (7)	

Abbreviations: CMH, Cochran–Mantel–Haenszel; HADS, Hospital Anxiety and Depression Scale; NGT, nasogastric feeding tube.

[a]Includes neurological/sensory conditions; heart/lung/immune system; skin/musculoskeletal conditions; metabolic conditions; abdominal conditions; kidney/bladder problems; and physical development of the eyes/ears/cheekbones/jaw/tongue/hands/feet/spine.

[b]*N* removed due to disclosure issues. Cells with <5 cases are not released in order to ensure that data remain non-disclosive.

authors using content analysis (Neuendorf, 2016). Common codes (in order of frequency, high to low) included feeding, speech development, sleep patterns, aggressive behaviours, vision, oral health, hearing, breathing and walking. Table 4 presents these codes alongside exemplar quotes.

4 | DISCUSSION

4.1 | Synthesis of findings and implications

This study assessed parent-reported socioemotional and cognitive development in children born with CL/P at 18 months of age. When compared with relevant cut-off scores, the sample as a whole indicated typical development across all subcategories. However, more in-depth analysis demonstrated that just over 31% of children in the sample met the criteria for onward referral in at least one domain, indicating a need for further assessment. A minority of children also met the criterion on three or more domains, highlighting potentially significant developmental difficulties that should be screened for by a clinician.

A number of factors provided evidence to suggest an association with the likelihood of a child meeting the referral criterion on at least one domain. First, almost 50% of children who needed additional NGT support and who also received formula milk met the criterion for at least one referral category. A study by Holden et al. (1997) found that the use of NGT was often very distressing for both parents and children, and as such, the authors advocated for adequate psychological preparation for families undergoing this medical intervention. Research also finds that tube feeding can be distressing for mothers, conflict with the development of a 'mother's identity' and potentially disturb the mother–child relationship (Wilken, 2012). As such, it is possible that the NGT experience during the child's first few days of life can have an adverse impact on the evolving child–parent relationship, subsequently affecting the child's developmental trajectory. Further, previous research has found that at 9 years old, children who were exclusively breastfed in the first three weeks of life experienced benefits in relation to their behavioural and cognitive development, likely due to the rich nutritious benefits of breast milk (Lanting, Fidler, Huisman, Touwen, & Bowersma, 1994). The World Health Organization (2013)recommends that all infants receive breastmilk exclusively up to 6 months of age to promote optimal physical and intellectual development. However, infants with CL/P are more likely to require feeding assistance, particularly if the palate is affected, as the provision of breast milk may not be possible (Cu & Sidman, 2011). However, there is also an ongoing debate about when NGT is indicated, with retrospective chart reviews and parent reports suggesting that children may be receiving NGT too hastily (Cu & Sidman, 2011). If NGT alongside formula milk cannot be avoided, early socioemotional and cognitive screening may be inferred.

The need for NGT and formula milk may also be indicative of physical abnormalities and/or a syndrome. Problems with the child's physical development were identified in the current study as being associated with at least one referral criterion. Physical anomalies in addition to CL/P could therefore indicate that further screening for socioemotional and cognitive development is necessary. Further, genetic testing is advocated in cases where developmental concerns are suspected (Stock, Zucchelli, Hudson, Kiff, & Hammond, 2019), and the ASQ could therefore be a useful tool in helping to identify those children who may be eligible.

Additional risk factors for developmental concerns were related to the demographic status of the mother. In a number of large-scale studies in the United Kingdom and internationally, children raised in single-parent families have been shown on average to have poorer outcomes than those whose parents are cohabiting or married (Waldfogel, Craigie, & Brooks-Gunn, 2010). This may be related to income and resource, parental involvement, parent–child relationships and parental well-being (Brody & Flor, 1997, 1998). Findings from the present study also suggest that single-parent families may require more support in coping with the additional challenges of having a child with CL/P, in order to enrich the child's development as well as parental well-being.

Child development was also associated with the mother's self-reported levels of anxiety and depression. Parental well-being has been shown to have a long-term impact on child outcomes in the general population (Sanger, Iles, Andrew, & Ramchandani, 2015), as well as in craniofacial populations (Pope, Tillman, & Snyder, 2005).

TABLE 4 Mother's qualitative reports of concerns for their child's development

Code	Exemplar quotes	*n*
Feeding	'Food and drink comes out of her nose. Cannot use a cup or straw'.	49
	'Will gag a lot and is sometimes sick'.	
	'Very fussy with food, doesn't like trying new textures'.	
	'Never seems hungry'.	
	'Eats excessively'.	
Speech development	'She is not making the sounds I feel she should be at 18 months old. No clear words'.	43
	'Delay in speech … mostly doesn't even babble'.	
Sleep patterns	'Has never had a full night's sleep'.	28
Aggressive behaviours	'Smacking, punching, biting'.	18
	'Head-butting and screaming'.	
Vision	'When he watches TV, he always sits very close to the screen'.	12
Oral health	'Teeth positioning, decay, future treatment, and development'.	9
Hearing	'Ear infections and impact on hearing'.	6
Breathing	'It's like something is blocking his breathing'.	4
Walking	'She is not yet standing without support or walking unaided'.	4

However, the literature also shows that having a child with healthcare needs (including CL/P) presents many challenges for parents, which can have a considerable impact on quality of life (Cousino & Hazen, 2013; Nelson et al., 2012; Smith, Cheater, & Bekker, 2015; Stock, Costa, White, & Rumsey, 2019). The cross-sectional nature of the current study resulted in an inability to identify the direction of this association. Nonetheless, previous studies have postulated that the relationship between parent and child adjustment is likely to be bidirectional (Pope et al., 2005; Smith et al., 2015). Overall, a healthcare approach, which implements both medical and systemic support for families from the point of diagnosis, is therefore advocated (Emerson & Bögels, 2017).

Content analysis of mother's qualitative data provided further insight into the condition-specific concerns that families may experience. These included weaning, sleeping, hearing and breathing difficulties, as well as concerns regarding vision and oral health. These are known challenges associated with CL/P (Richman et al., 2012; Stock & Feragen, 2016) yet may not otherwise be picked up at this early stage depending on the recommended treatment pathway.

4.2 | Utility of measures

The ASQs appear to offer a viable option in the identification of parent-reported developmental concerns in the CL/P population and could potentially be extended to the wider craniofacial population. The measures are relatively brief and are completed by the parent, reducing clinical assessment time and drawing upon parent expertise. As an initial screen with the potential to identify the children in need of further assessment, the ASQ also offers an appealing alternative to the time-consuming battery of tests that are currently recommended in some countries (Collett & Speltz, 2006). Versions of the ASQ are available from the age of 1 month to 5.5 years, and children's progress can therefore be followed longitudinally. The qualitative component offers an opportunity to identify condition-specific concerns based on parents' observations and to begin a dialogue about these concerns. Teams working with children with craniofacial conditions and their families could adapt the ASQ measures as a way to review the child's development. If the ASQ measures are already given out as part of routine assessment, craniofacial teams should be given access to that information in order to avoid data duplication and reduce the burden on parents.

4.3 | Methodological considerations

Comprehensive data were extracted from The Cleft Collective Birth Cohort Study (Stock 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 mothers, with those from Black and minority ethnic communities, those having immigrated to the United Kingdom and those with lower socio-economic status are under-represented 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 under-representation 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 subgroups. Further work is needed to ensure that studies are relevant and accessible to all eligible participants.

It is important to acknowledge that the current findings likely apply to the general population and may not be unique to children with a CL/P. Indeed, the use of formula milk and/or NGT, the presence of physical anomalies in the child, decreased maternal well-being and living in a single-parent household are factors increasingly associated with affecting normal child development. However, the literature suggests that in comparison with children from the general population, those with health conditions, like CL/P, are at an additional risk of experiencing a range of developmental concerns (Richman et al., 2012; Stock & Feragen, 2016). Therefore, it is important to study samples who are at a likely greater risk of challenges in order to prevent long-term disadvantage.

The findings of the present study also demonstrate the utility of the ASQ in craniofacial research. However, it is important to acknowledge that parent-reported concerns are not necessarily always indicative of actual developmental delay or disability, particularly at 18 months when 'normal' development varies considerably. Indeed, although parents know their children well, parent perceptions may be strongly influenced by other issues such as their own well-being. As such, the use of the ASQ should always be premised with the intention to follow-up with clinical assessment as necessary.

Last, this study only sought to explore the development of children at 18 months. However, many of the longer term concerns that can impact children with CL/P do not become evident until further in the child's life, for example, in relation to academic outcomes (Knight et al., 2015; Wehby et al., 2014). As such, continued screening at different developmental stages is indicated. The ASQs can be used as screening tools for children between the ages of 1 month and 5.5 years; however, beyond that, other tools, such as the Strengths and Difficulties Questionnaire (Goodman, 2001) should be implemented.

5 | CONCLUSIONS

The current study assessed the prevalence of parent-reported developmental concerns in children with CL/P at 18 months, as well as associated factors. The ASQs appear to offer a viable option in the early identification of parent-reported developmental concerns in the CL/P population. A combined medical and systemic approach to healthcare is recommended to support the prevention of long-term developmental concerns in the child and poor psychological adjustment in parents.

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CONFLICT OF INTEREST

The authors have no disclosures to make.

ADDITIONAL MATERIALS

For any additional materials associated with this manuscript, please contact the corresponding author.

ORCID

Bruna Costa https://orcid.org/0000-0001-8275-695X

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