**Caring for a Child with a Cleft Lip and/or Palate: A Narrative Review**

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**Abstract**

Raising a child with healthcare needs places additional demands on caregivers. In 2012, Nelson and colleagues authored a review of 57 papers pertaining to parents’ experiences of caring for a child with cleft lip and/or palate (CL/P). Thanks in large part to this review, available literature on this topic has grown considerably. The aim of the present review was to update and critically appraise recent literature, with the wider goal of assessing progress in the field and setting recommendations for future work. All original, peer-reviewed articles pertaining to the psychological adjustment of parents of children with CL/P living in high-income countries (published May 2009 to May 2024) were examined. A total of 126 articles were included. Findings were narratively synthesised according to three salient themes: Emotional Impact; Social Experiences; and Care Delivery. Recent research has built on Nelson et al.’s recommendations, addressing some prior gaps in knowledge. Nonetheless, some areas remained largely unexplored and critical methodological limitations were still evident. Recommendations for clinical practice include: improved informational resources for parents and non-specialist health professionals, regular audit of services in collaboration with parents and families, routine psychological screening for known risk factors and integrated psychological support from diagnosis onward. Recommendations for future research include the design of multicentre, prospective, longitudinal studies with sufficient sample sizes and appropriate control/reference groups, inclusion of families from diverse ethnic and socioeconomic backgrounds, further examination of factors contributing to psychological growth, the development and evaluation of psychological interventions, and cross-condition learning.

**Key words:** cleft lip and palate; parent; caregiver; quality of life; mental health; social support; treatment

**Introduction**

Raising a child with healthcare needs represents a significant additional demand on caregivers and families1-3 (Cousino & Hazen, 2013; Cohn et al., 2020; Bayer et al., 2021). Stressors commonly relate to the child’s physical health and developmental needs, treatment decision-making, and socioemotional challenges, alongside a variety of practical considerations, such as an impact on finances and employment, accessing appropriate services, the volume and location of appointments, and the psychological wellbeing of the wider family unit1,4-5 (Melnyck et al., 2001; Cousino & Hazen, 2013; Masefield et al., 2020). Rather than representing a singular event, chronic medical conditions typically initiate a series of events in which parents experience various highs and lows, interact with a myriad of healthcare providers and systems, and must learn to manage their child’s fluctuating healthcare needs6.

Cleft lip and/or palate (CL/P) is one of the most common congenital conditions in the world, affecting 1 in 1,000–1,500 live births globally7 (World Health Organization, 2023). While primary surgery to close the lip and/or palate normally occurs during the first year of life, ongoing multidisciplinary treatment is typically required8 (Hodgkinson et al., 2005). Following an antenatal or postnatal diagnosis of CL/P in their child, caregivers must therefore adjust to the implications of their child’s condition and embark on a long-term treatment pathway.

In 2012, Nelson and colleagues authored a review of 57 papers published between 1980 and 2009 pertaining to parents’ experiences of caring for a child with CL/P9 (Nelson et al., 2012). Bringing quantitative and qualitative literature on this topic together for the first time, the review identified a series of salient themes. The first theme focused on parents’ emotional experiences of having a child with CL/P, in which parents reported a broad range of emotional responses to their child’s diagnosis, an emotional impact of feeding difficulties, and elevated stress and reduced mental health. Some studies also explored the potential impact of CL/P on parent-infant bonding and a variety of different coping strategies utilised by parents. The second theme highlighted the impact of CL/P on families’ social experiences, including perceived stigma, social exclusion and social support. The third and final theme explored parents’ experiences of CL/P services, with a focus on the notable dearth of reliable information about the aetiology and prognosis of CL/P, as well as a perceived burden of care, a lack of involvement in treatment decision-making, and concerns about access to and overall coordination of CL/P services.

Moving beyond the CL/P literature, Nelson and colleagues compared the 57 CL/P papers to broader literature on long-term conditions9. Clear similarities in the social, emotional, and service-related experiences of parents were found across conditions. However, the authors critiqued the CL/P literature for its exclusive focus on parents’ (predominantly mothers’) experiences of the early years, and the comparative lack of exploration of broader holistic approaches and theories. Methodological constraints were also clearly identified, including an emphasis on deficit-oriented approaches, a reliance on cross-sectional methodology with small sample sizes, and a relative paucity of qualitative research.

The review by Nelson and colleagues has been widely cited and commended, yet it is now more than 10 years old. Thanks in large part to the review, many researchers around the world have since focused their efforts on examining parental well-being in relation to having a child with CL/P. The aim of the present review was to provide an update and critical appraisal of the literature published since Nelson et al.’s original review was completed, with the wider goal of assessing progress in the field and setting recommendations for future work.

**Methods**

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Checklist (2020)10 was followed where applicable to ensure the quality of reporting. This included examination of the review against the PRISMA 2020 Item Checklist and the inclusion of a flow diagram.

***Inclusion Criteria***

All original, peer-reviewed articles pertaining to the psychological adjustment of parents of children born with CL/P published between May 2009 (based on Nelson et al.’s 2012 inclusion report)9 and May 2024 were included. Quantitative, qualitative, and mixed-methods papers were considered. Articles relating to all types of syndromic and non-syndromic CL/P were included. No age restrictions for participants or their children were enforced. Articles published online while ‘in press’ were also included where available. Articles published in all languages were included where English translations could be reliably obtained.

***Exclusion Criteria***

Case studies, protocol papers and unpublished dissertations were excluded. Articles relating to ‘visible difference’, ‘disfigurement’, ‘craniofacial conditions’, or similar were excluded where results were not separated according to condition. Articles describing findings from low- and middle-income countries (as defined by the World Bank Classification) were excluded, as these are the focus of a separate review (*article in preparation*). No literature reviews, systematic reviews, summary articles, book chapters or meta-analyses published during the search period were included but were stored separately for reference.

***Search Strategy***

The current review used a similar but more focused search strategy compared to the one carried out by Nelson et al9. Rather than reviewing all literature pertaining to long-term conditions, this review concentrated on CL/P specifically. Databases included PsychInfo, MEDLINE, CINAHL Plus and Scopus. Search terms identified within the article title, abstract or keywords included parent\* OR mother\* OR father\* OR caregiv\* OR famil\* OR maternal AND cleft OR cleft lip OR cleft palate OR orofacial cleft OR craniofacial AND emotion\* OR social\* OR psych\* OR wellbeing OR well-being OR adjust\* OR quality of life OR stress OR depress\* OR anx\* OR stigma\* OR cop\* OR distress OR resilien\*. The reference lists of previous reviews were hand-checked to reduce the likelihood of any abstracts being missed. Any duplicates were removed. Titles and abstracts were screened by two independent reviewers. To assess quality control, 40% of abstracts were double screened. The agreement rating was 98.3 percent (Cohen’s kappa: 0.79). Any minor discrepancies were discussed until full agreement was reached. Full texts were then screened by the first author (Figure 1). Data regarding methodological details and key findings were extracted from each included paper by two reviewers and cross-checked for accuracy.

**Results**

In total, 126 articles met the inclusion criteria. Three overarching, novel themes were identified: Emotional Impact; Social Experiences; and Care Delivery. Data extraction for each article is provided in Table 1. Results are narratively synthesised below according to each theme and associated subthemes.

***Narrative Synthesis***

1. **Emotional Impact**

Seventy-two papers reported on the emotional impact of having a child with CL/P. Subthemes included parental reactions to the diagnosis (24 papers), common parental concerns (22 papers), the emotional impact of feeding difficulties (7 papers), parent-infant bonding (12 papers), the impact of additional conditions/syndromes (9 papers), parental mental health (29 papers) and parents’ coping strategies (11 papers).

* 1. *Parental Reactions to the Diagnosis*

A wide range of emotional reactions to the diagnosis was reported by parents. This included shock, worry, sadness, overwhelm, depression, guilt, grief, panic, heartbreak, confusion, fear, despair, anger and a sense of unfairness, alongside concern for the future11-18. Some parents also expressed delight13, while others felt the joy of having a baby had been somewhat diminished by the diagnosis11,19. While some parents didn’t feel that CL/P was a major concern, or believed cleft lip was mostly a cosmetic issue that could be addressed15,20-22, many grappled significantly with definitions of normality, perfection and difference13,18. Those parents that perceived CL/P to be a more significant health condition or disability reported a greater anticipated impact on their own and their child’s future happiness20. Rates of antenatal diagnosis varied according to country and methodology, but overall prenatal detection rates of cleft lip were high. While some papers found no differences in parental wellbeing in relation to the timing of the diagnosis12,23, others identified greater concerns in those receiving a diagnosis postnatally15,17,24. Parents’ acceptance of their child’s appearance was also more negatively impacted if they received the diagnosis after birth17. In contrast, receiving a prenatal diagnosis of CL/P gave parents more time to adjust and prepare11,15,18,25. Detection rates of cleft palate only were more variable, with some reports of delayed diagnosis resulting in parental distress15,26-27. A lack of understanding of the aetiology of CL/P was associated with greater self-blame in parents12, which in turn predicted poorer general well-being, depression, elevated stress and anxiety scores, and a negative impact on parent-infant bonding28-31.

* 1. *Common Parental Concerns*

Parents identified a series of concerns related to their child’s CL/P. These concerns typically arose following a diagnosis but were still evident in samples of parents of older children. The most common concerns centred on the child being susceptible to teasing, stigma and poorer emotional health due to appearance- and/or speech-related differences9,11,15,17-18,22,32-37. Other concerns included the presence of additional conditions or syndromes, aetiology, managing feeding difficulties, explaining CL/P to others, dealing with other people’s reactions, the burden of care and the recurrence risk of CL/P11-12,14-15,18-19,21-22,25,36,38-41.

* 1. *Feeding Difficulties*

Feeding difficulties were the cause of frustration, anxiety, distress, sadness and despair for parents14,26,42. Some mothers also described a sense of loss at not being able to breastfeed their child as they had planned18. Feeding difficulties were described by mothers as traumatic in one study41, while in two others, parents reported feeling personally responsible for their child’s failure to thrive27,43. Using breast pumps was considered to be stressful and time-consuming14. Problems with feeding and a longer duration of feeds predicted poorer maternal health-related quality of life in two studies43-44.

* 1. *Parent-Infant Bonding*

Considerable variations were observed across studies of parent-infant bonding. Two studies found no differences in maternal representations or parent-reported attachment quality between mothers of children with CL/P and those without45-46. In contrast, three other studies identified significantly more disengaged attachment representations and negative interaction patterns exhibited by mothers of children with CL/P47-49. One study found that mothers of infants with cleft lip gazed at their infants’ faces less often than controls50. Mothers also reported bonding insecurities, concerns about a lack of physical closeness and reduced maternal instinct14,41. In one experimental study, Gassling and colleagues found parents of children with CL/P to be more encouraging and less directive and restrained compared to controls, which led to the child exhibiting greater independence and initiative51. Parents qualitatively reported no impact and/or a positive impact on bonding in one study exploring parents’ early experiences52. Nasoalveolar Moulding (NAM) treatment was found to both positively and negatively impact bonding according to parent report53.

* 1. *Additional Conditions*

Parents described the emotional impact of a range of additional conditions and symptoms related to CL/P. In a study by Tierney et al54., parents reported feeling frustrated with their child’s inability to hear well and the subsequent impact on their child’s behaviour and learning. Parents also experienced anxiety around hearing appointments, feeling helpless with regard to frequent ear infections, and needing to act as an intermediary when their child was struggling to communicate54. Berger and Dalton found hearing difficulties to account for 6.6% of the variance in parental wellbeing28. In two other qualitative studies, parents expressed frustration at their child not being able to communicate intelligibly with others15,41. Speech was a particular challenge for parents of internationally adopted children, given the language barrier33,55. Parents of children with a syndrome reported more impact on parent and family well-being and greater levels of stress, post-traumatic stress, hostility and depression56-59.

* 1. *Parental Mental Health*

Findings in relation to parent mental health were highly variable depending on the sample and outcome measures used. Some studies found no differences between parents of children with CL/P and control/reference groups in relation to depression, anxiety, stress, post-traumatic stress symptoms or overall quality of life23,48,58-64. In contrast, other studies identified elevated stress, anxiety, and depression, alongside more symptoms of emotional difficulties, post-traumatic stress and decreased quality of life23,35,47,63,65-70. Three studies identified elevated postnatal depression scores in mothers24,43,71, with 11.7 percent of mothers reporting scores above the clinical cutoff in one study24. Mothers in this sample also reported feeling anxious, scared and sad, with 1.5% endorsing minor intent for self-harm24. Despite elevated anxiety and depression scores compared to a normative sample, scores remained in the normal range in two studies66,71. Parents qualitatively described elevated levels of anxiety during the Covid-19 pandemic72. Other studies reported lower levels of stress, less anxiety, and better overall quality of life than control/reference groups35,59,66. Stress scores fluctuated according to treatment stage in one study35. Predictors of poorer mental health in parents of children with CL/P included lower annual income, fewer years of parental education, having a greater number of children, behavioural problems in the child, parents’ older age, the presence of a prior mental or physical health condition, prior stressful life events, and unexpected absences from work24,28,59,62,66,73-74. A positive life orientation was identified to protect parents from psychological distress66. Five studies found no variations in parental well-being according to cleft type23,37,59,66,75, while others identified a higher impact of cleft lip and palate compared to cleft lip or cleft palate only37,65,68,73,76, or a reduced impact for those with cleft lip only58,77. Mothers scored significantly less favourably than fathers on measures of mental health in some studies66,73. Psychological distress was found to reduce over time in two studies63,77. Parents in two studies specifically identified a need for dedicated emotional support12,13.

* 1. *Parents’ Coping Strategies*

Parents described a range of emotion-focused, problem-seeking and meaning-making coping strategies, in addition to seeking social support and support from health professionals. Specific coping strategies included adopting an optimistic outlook, making downward comparisons, drawing on faith, problem-solving, leaving the house less often, avoiding taking photographs of the baby, engaging in own research and advocacy14,18,35,53,78. Active coping and seeking emotional and social support were associated with family resiliency in one study31. Coping strategies associated with lower resiliency and lower positive affect included restraint coping, substance use, self-blame and denial31. Personal growth was facilitated through parents and couples taking an active role in the treatment process, putting CL/P into perspective, recognising strengths, and reinforced relationships15,22,29,53,79-80.

1. **Social Experiences**

Thirty-two papers reported on parents’ social experiences in relation to having a child with CL/P. Subthemes included other people’s reactions (6 papers), the marital relationship (6 papers), family functioning (6 papers) and social support (22 papers).

* 1. *Other People’s Reactions*

Several papers highlighted the stigmatising reactions reported by parents in relation to their child’s CL/P. These reactions could be from friends, family members, and members of the public, as well as health professionals13,22,33,36,52. Curiosity from strangers and comments about their child’s appearance were often experienced by parents as hurtful or distressing and could have a lasting impact13,22,36,52,57. Some parents felt vulnerable to strangers’ comments and therefore chose to hide the child while outside of the home, or to stay in the house to protect themselves18,36. Parents chose not to share photographs of their child prior to lip surgery in one study18. Some comments were perceived by parents to be more neutral or positive in nature17-18,36, although well-intended comments could also be seen as trivialising36. Whispers, stares and pity were also reported36. Some parents felt irritated or angry, while others were happy to be asked about their child’s CL/P and felt an open, calm and practical approach was most helpful in dealing with comments36.

* 1. *Marital Relationship*

One study by Maarse and colleagues20 demonstrated that parents’ appraisals of their child’s CL/P were strongly influenced by their partner’s reactions to the diagnosis. When partners responded positively, the negative impact of the diagnosis was lessened81. Stock et al. also found relationship satisfaction to be protective against psychological distress for both mothers and fathers66. In contrast, having a child with CL/P was found to change the couples’ relationship and/or be the source of marital discord in some cases16,19. Yet, if the couple were able to work together in times of stress, the marital relationship could grow in strength14,16,1,9,41.

* 1. *Family Functioning*

A handful of studies investigated the impact of CL/P on the psychological health of the family unit more broadly. This included a negative impact on the quality of interaction between family members, a reduction in family activities and an increase in the degree of family conflict65,67,82. A total of 36.5% reported ‘a lot’ or ‘some’ impact of CL/P on family life in a study by Agnew and colleagues75. However, in two multicentre studies with >1,000 participants, scores of family cohesion, expressiveness, conflict were in the normal range83 and both mothers and fathers reported more favourable scores for daily activities and family relationships compared to normative data66. Sischo and colleagues also identified greater family expressiveness and less conflict in families with CL/P compared to published norms35. A range of predictor variables were identified in relation to family impact, including a prior mental health condition in the parent, degree of clinical need, treatment stage, type of health insurance, sociocultural variables, a positive life orientation, healthcare satisfaction and relationship satisfaction35,66,83.

* 1. *Social Support*

Several papers noted the importance of family support for parental adjustment to CL/P. This support could be emotional or practical in nature14,36,44,57. Utilising family support was seen as a core coping strategy in some studies18,11,16,35,41, while unsupportive comments or behaviours by family members were harmful to parental well-being22,36. Similarly, changes in friendships could be hurtful for parents36, while close friendships were found to be protective against depression in mothers66. Parents also sought support from peers via non-profit organisations, social media and hospital-based support groups. Accessing peer support was also viewed as an important coping strategy, with parents citing the opportunity to share their experiences with others, feeling less alone, reassured and more able to cope with cleft-related challenges11,14,18,22,35,53,84-86. Some non-profit organisations also provided physical resources such as feeding bottles and information leaflets, which parents found helpful16. In other cases, parents identified a lack of social support and wanted information about and access to support organisations and local parent groups13,38,87. A large proportion of parents had also used the internet, including social media to access information about CL/P22,40,88. One study found a perceived lack of social support to predict depression, anxiety, less self-control and poorer well-being and vitality29, while another found effective social support to be correlated with resiliency31.

1. **Care Delivery**

Eighty-five papers reported on parents’ experiences of CL/P care. Subthemes included the diagnostic experience (33 papers), feeding support (10 papers), treatment decision-making (17 papers), treatment experiences (34 papers) and healthcare satisfaction (35 papers).

* 1. *Diagnostic Experience*

Whether diagnosed prenatally or after birth, parents frequently reported non-specialist health professionals, such as sonographers, midwives and paediatricians to lack knowledge of CL/P, which in turn had a negative impact on their appraisals of what CL/P would mean for their family15,17-18,20-22,25,27,38,41. Many parents receiving a diagnosis antenatally had been asked to consider terminating the pregnancy without an opportunity to fully understand the diagnosis20,22,26,89, while others had felt pressured to undergo amniocentesis with a view to terminating the pregnancy if a positive result was identified11,89. Those parents that came close to ending their pregnancy reported significant distress20,89. After the birth, health professionals’ lack of knowledge could lead to delayed diagnosis, parents spending less time with their baby and delayed referrals to specialist teams26-27. A number of papers also described unhelpful, insensitive or dismissive reactions from non-specialist health professionals that had a lasting impact on parental well-being15-16,18,21-22,26-27,41,89. Parents of adopted children had not always been told about their child’s medical status prior to adoption, and those that had felt this information was inadequate33. In response to a lack of sufficient information, parents often utilised the internet and social media to learn more about CL/P and its treatment11,13,16,21-22,27,38, yet the quality of online information was found to be highly variable17,18,39-40,88,90-99. Parents wanted reassurance from health professionals, counselling opportunities and consistent and accurate information to reduce their anxiety11-12,20,25,38. Families who were followed up early by a specialist nurse were less likely to utilise online support in one study64. Differences in the desired level and timing of information were observed11,34,38,100, with some parents preferring not to view pre- and post-surgery photographs16,38-39. On the whole, parents felt reassured and much more informed once under the care of a specialist health professional or team22,38,93,101.

* 1. *Feeding Support*

Information for parents about how to feed a child with CL/P was reported to be inadequate across a number of studies, which resulted in parents feeling anxious and unprepared11,14,87. Non-specialist health professionals were also perceived to lack the expertise necessary to meet the child’s feeding needs14,41,102. A large proportion of parents received no encouragement to breastfeed or were actively discouraged according to two studies74,87, and rates of continuation fell behind the national average44,74. Despite mothers being motivated to breastfeed, challenges such as feeding being too complicated, too stressful, too difficult, too time-consuming and too painful were cited as key reasons for discontinuation74. Yet, when parents were provided with specialist information, counselling, individualised lactation support and practical guidance, rates of continued breastfeeding were high12,14,86,97. Additional barriers to establishing an effective feeding routine included difficulty obtaining a specialist feeding bottle, the cost of bottles and needing to try several bottles before achieving some success87.

* 1. *Treatment Decision-Making*

A common desire among parents was to do the ‘right’ thing by their child. For many, this involved accessing all the treatment available32,103. The pursuit of treatment was also driven by a need to reduce the likelihood of real or anticipated social stigma13,15,32. Yet, parents also reported conflicting feelings about sanctioning treatment, particularly if the primary goal was to ‘normalise’ their child’s appearance18,22,32,35. Parents worried about the risks of surgery, pain management and the physical and emotional impact of treatment on their child11,18,35,55,104-105. Trust in the medical team was therefore essential, and many parents chose to follow health professionals’ treatment recommendations11,13,15,19,22,32. There was little evidence of shared decision-making, with parents not always being given the range of options available, not fully understanding the treatment process and/or feeling coerced into choosing a particular option16,106-107. One study reported that parents wanted to be involved in decisions and to take a proactive role in their child’s treatment108. Yet, minimal agreement between parents and children about proposed treatment plans was highlighted, with some parents only allowing their child responsibility for treatment decisions as they got older32,103,109.

* 1. *Treatment Experiences*

Parents described the burden of CL/P care, including financial burden, the frequency of appointments, impact on employment, lack of care coordination, childcare difficulties, long wait times and distance travelled11,18,22,34,56,104-105,108,110-114. This burden was greatest for parents with less education, parents belonging to an ethnic minority group and parents speaking languages other than English56,111,115. A greater impact of treatment predicted poorer global well-being, vitality and general health, and greater anxiety in one study29. Long-term treatment involved peaks and troughs, periods of stability, intensive stages of treatment, periods of waiting to see the results, exhaustively advocating for the family’s needs to be met and dealing with ongoing uncertainty13,22,41,53. Having continuous access to a highly experienced and specialist team alleviated some concerns and produced better parent-reported outcomes11,18,22,26,34,102,104,108,116-117, yet team intervention was still described by some parents as overwhelming22,80,102. Surgery was seen as a major stressor for parents, particularly if cancellations occurred, the benefits of treatment were not immediately obvious, there were unexpected complications, or if a surgery needed to be repeated13,19,22,54,106,118-119. Some parents stated they had not been prepared for the change in their child’s appearance after surgery and had found this distressing11,22. Three papers described parents’ traumatic stress reactions to medical treatment, which was worsened by parents’ perceptions that they were not being believed or listened to by health professionals41,54,107. Five studies specifically investigated parents’ experiences of NAM. Some parents reported that NAM became less stressful and more empowering over time as they began to master the process35,53,91. Reasons for discontinuation of NAM included sleep apnoea in the child, device intolerance, issues with taping and a lack of support, as well as the mother being younger, being a single parent, having longer travel distances to the hospital and having less insurance coverage120-121. Parents who did not complete NAM were less satisfied with the outcome of surgery121. Parents described needing to see measurable progress to support the efficacy of treatments and to make the burden of treatment worthwhile41,80,104,106,108.

* 1. *Healthcare Satisfaction*

Overall, parents reported a high level of satisfaction with the care they and their child had received from the specialist CL/P team22,33-34,39,56,64,102,104-105,100-111,117,122-123. Parents particularly valued professionals’ knowledge and technical competence, professionals’ interpersonal skills and continuity of care, as well as repetition of information, access to psychological support and reassurance about their child’s development25,104,124. Some unmet needs were identified by parents, including a desire for more written information on a wider range of topics, information and support tailored to the family, training for non-specialist health professionals, increased contact with other parents and more consistency of information and communication between health professionals25,104,124. Fathers were found to be particularly dissatisfied with their access to credible information and their inclusion in medical appointments and support networks in four studies22,64,73,123, which worsened during the Covid-19 pandemic72. During the pandemic, reduced contact with health professionals, changes to surgical protocols and surgical delays negatively impacted parents’ experiences of care72,125. However, access to telehealth was broadly viewed as a potentially useful adjunct for future in-person care72,125,126. Eight studies specifically described parental satisfaction with the aesthetic and functional outcomes of NAM35,53,102,127 and surgery11,15,52,81. However, a small proportion of parents reported less favourable perceptions of treatment outcomes, including the appearance of their child’s nose, lip and teeth, and dissatisfaction with their child’s ability to hear and breathe well42,78,81-82,115,128-130. Parents were less satisfied with treatment if the child had combined cleft lip and palate82,111,115. Parents and children were prone to disagreement, with parents being less satisfied with treatment outcomes and having a greater desire to pursue further surgery81,128-131.

**Discussion**

*Data Synthesis and Comparison to Nelson et al. (2012)*

This narrative review synthesises the literature published on parents’ experiences of caring for a child with CL/P since Nelson et al.’s original review was completed in 20099. Three overarching, novel subthemes were identified: Emotional Impact, Social Experiences, and Care Delivery. These themes are similar to those presented in Nelson et al.’s 2012 review9.

As identified in Nelson et al.’s 2012 review9, parents reported a wide range of emotional responses to their child’s diagnosis of CL/P, followed by a series of common concerns relating to the aetiology of CL/P, the treatment pathway and the long-term impact of CL/P on the child and wider family unit. Parents across both reviews struggled to access reliable information from non-specialist health professionals at the time of diagnosis, turning instead to internet sources of variable quality. What is novel, however, is an improved understanding of how parents’ early interactions with health professionals and initial degree of CL/P knowledge impact on their response to the diagnosis, the choices they make about antenatal testing and termination and their appraisals of what CL/P may mean for their family. Parents with more negative appraisals of CL/P, internal attributions of the cause of their child’s condition and/or unmet information needs are more likely to experience poor mental health and a greater impact on familial relationships. The timing of the diagnosis may also play a role, in that a diagnosis during pregnancy gives the parents time to adjust and prepare before the baby arrives.

Once the baby has been born, parents’ attention typically focuses on feeding. The emotional impact of feeding difficulties has been explored in more depth since Nelson and colleagues published their review. Parents who lacked information, encouragement and practical support to feed their baby were less likely to breastfeed or continue breastfeeding, and were more likely to experience grief, guilt, bonding insecurities and poorer quality of life. Yet, rates of breastfeeding were comparable with the national average when specialist feeding advice and equipment was provided.

Many more studies have examined parental mental health and family functioning since the original review was published. While findings remain highly variable, a marked impact of CL/P on parental and familial well-being is evident in a subsample of vulnerable families. Importantly, studies have begun to identify predictive factors for psychological distress (Table 2), such that identification of parents who may be at risk has been made more possible. The presence of a syndrome and its impact on parental well-being has also been more readily discussed in recent literature, indicating research may have become more inclusive of all cleft types132. Another welcome contribution is the inclusion of the father’s voice in CL/P research, and while it seems fathers may be less emotionally impacted by their child’s diagnosis than mothers overall, research has identified a gap in support tailored to fathers’ specific needs. While examination of coping strategies, personal growth and resiliency remains relatively scarce, an increased interest in the factors that protect parents from distress is a welcome contribution to current knowledge.

As described in Nelson et al.’s review9, other people play a key role in how parents adjust to the news of their child’s condition. Recent research has confirmed the emotional impact on parents of hurtful reactions from strangers, explaining in more detail the strategies parents use to cope with this. These societal perceptions, alongside the opinions of friends and family members were found to disrupt or facilitate parents’ adjustment to the diagnosis and influence parents’ motivations for treatment through the introduction of constructs such as ‘abnormality’ and ‘difference’. Another aspect not previously documented is the significant influence of romantic partners on how parents perceive CL/P and how in turn, the ability of couples to work together to cope with CL/P-related challenges can impact individual well-being and the marital relationship. Building on Nelson et al.’s suggestion that support from significant others may be important for parental adjustment9, the current review identified social support as a key coping strategy which can take many forms, including connecting with other families affected by CL/P.

In line with Nelson and colleagues’ original review9, overall satisfaction with specialist CL/P services was found to be high. Yet, the burden of ongoing care, poor care coordination and communication between health professionals, pre- and post-operative challenges and inadequate quality and delivery of information continued to impact parent well-being. A lack of shared decision-making was also indicated, which could impact parents’ satisfaction with treatment outcomes and, in some cases, could evoke traumatic stress reactions. Parents’ desired level and timing of information and degree of involvement in treatment decisions was found to vary considerably, emphasising a need to understand how to tailor information and support to the needs of individual families.

*Implications for Clinical Practice*

Taken together, the findings of the current review highlight a number of considerations for clinical practice (Table 3). Improvements in the quality, relevance, and accessibility of CL/P-related information for both parents and non-specialist health professionals is recommended, to reduce the risk of misinformation and moderate the emotional impact of the diagnosis. Specialist feeding advice and equipment could also minimise parental distress and enhance parent-infant bonding, while ensuring the infant is receiving adequate nutrition. Involvement of the father/partner in healthcare appointments may help them to feel included in decisions about their child’s health, as well as provide opportunities to assess their well-being and the health of the marital relationship. In the absence of a robust evidence-base for intervention133, brief psychoeducational resources may be effective in alleviating common concerns and in encouraging parents to seek further psychological support if needed. Similarly, facilitating peer support opportunities for families can be a powerful tool134. Including a psychosocial specialist on all CL/P teams and ensuring they are visible and accessible to families from the diagnosis onward is also crucial for identifying and addressing any concerns, as is the consistent use of appropriate screening tools. CL/P teams may also benefit from regular audit of their services in collaboration with parents to identify areas of strength in service delivery and opportunities for improvement.

Despite the notable growth in our understanding of parental concerns, integration of psychological support and resources for affected families into the routine treatment pathway is still not commonplace in paediatric care, and neither is screening to identify parents and families in need of support1. Preventing and addressing mental health concerns in parents and families should be a priority to ensure the well-being of individual parents, the affected child and the wider family unit.

*Implications for Future Research*

In comparison to the original review, which accounted for 57 papers published over the course of 29 years (1980-2009)9, a far greater number of eligible papers were identified in the current review (between 2009-2024; 15 years; *n*=126). In response to Nelson et al.’s recommendations9, an increase in the number of qualitative studies is notable. Unfortunately, the lack of exploration of parents’ experiences of the later years (mid-childhood onwards) and use of broader holistic approaches and theories remains stark. Models from the wider health field, such as parental stress and coping in the context of chronic illness and/or disability135-138, could have utility in craniofacial research and practice. An emphasis on single-centre, cross-sectional, deficit-oriented studies utilising small sample sizes without control/reference groups persists, as does an inconsistent use of (often unvalidated) outcome measures139-140. Few studies include adequate numbers of parents from ethnic minority communities and/or low socioeconomic backgrounds to fully understand the needs of vulnerable subgroups. Specific topics that warrant further research exploration according to the findings of this review include an understanding of the type of information families need at each stage, examination of the factors that contribute to personal growth, and additional research into effective shared decision-making. In addition, few intervention studies were identified in the current review, emphasising the ongoing need to develop and assess psychological interventions in this field133,141. Given the similarities between the psychological and healthcare experiences and needs of parents of children with CL/P and those impacted by other long-term health conditions1-5, cross-condition learning could be highly beneficial in moving the field forward. Recommendations for future research are provided in Table 3.

**Conclusions**

This narrative review of recent literature has confirmed a broadly adequate depth of understanding of the challenges experienced by parents of children born with CL/P. An important next step will be to move away from simplistic descriptions of parents’ experiences and toward a more complex assessment of how clinical teams can best facilitate psychological adjustment and personal growth.

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**Figure 1:** Flow Chart Demonstrating the Selection of Articles for Inclusion

A screenshot of a computer

Description automatically generated

**Table 1:** Data Extraction Table

|  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Author(s) and year** | **Recruitment site(s)** | **Child’s diagnosis** | **Child’s age range** | **Exclusion criteria** | **Sample size** | **Measurement** | **Comparison group** | **Theme(s)** |
| Acharya, 2022 | Single centre (USA) | CL or CP | ≤ 28  ≥ 37 weeks | Non–English-speaking, non-biological parents, infants previously discharged, infants transferred to cardiac intensive care, infants for whom death was imminent | Baseline: 166 parent-infant dyads  Follow-up: 124 parent-infant dyads  Cleft: 12 parent-infant dyads | Pediatric Quality of Life (Family Impact Module) PedsQL-FIM | Published data on other major congenital anomalies | Emotional |
| Agnew et al., 2020 | Single centre (Australia) | CL / CP / CLP / VPI / submucous cleft | 7-18 years | Child with a known syndrome | 214  (129 mothers, 85 fathers) | Family Impact Scale Short-Form (FIS-SF) | None | Social |
| Akiki et al., 2021 | GoFundMe website  (USA) | CL, CP, CLP | N/A | Duplicate campaigns, campaigns outside the USA, campaigns created by an organization, raising funds for a pet rather than a child | 635 crowd-funding campaigns | Google Trends, with values reported as Relative Search Volumes (RSV), Google Maps data | Within-group comparisons | Care |
| Alfonso et al., 2024 | Single centre (USA) | CLP | Supplementary material only | Syndromic facial clefts, NAM therapy performed at other institutions, initial presentation for revision surgery, presurgical assessment unavailable | 230 patients | Medical records | Within-group comparisons | Care |
| Alighieri et al., 2020 | Single centre (Belgium) | CP / CLP | 5-13 years | Parents with craniofacial anomaly themselves | 11  (5 mothers, 6 fathers) | Semi-structured interviews | None | Care |
| Alighieri, 2021 | Single centre (Belgium) | CL / CP / CLP | 6-10 years | Cognitive/related learning disabilities or syndrome, oronasal fistula, VPI, hearing difficulties | 12 mothers | Non-validated questionnaire and semi-structured interviews | Within-group comparisons | Care |
| Alighieri, 2023 | Single-centre (Belgium) | CL / CP / CLP | 6-10 years | Not reported | 7 mothers | Semi-structured interviews | Within-group comparisons | Care |
| Al-Taha, 2019 | Single-centre (Canada) | CP | 9-11 months | Lack of English fluency; inability to read at a grade 7 level; discussion of additional procedures; inability to provide informed consent | 40 parents | Semi-structured interview | Within-group comparisons | Care |
| Antonarakis & Kiliaridis, 2009 | Internet search  (Global) | CL / CP / CLP | N/A | N/A | 49 websites | Flesch Reading Ease Score / Flesch-Kincaid Grade Level | N/A | Care |
| Arslan, 2024 | YouTube videos  (Global) | CL / CP / CLP | N/A | Surgery videos, videos using a technique other than NAM,  content not in English, videos not  related to the subject | 24 videos evaluated | Interaction index, viewing rate, Global Quality Scale (GQS), content evaluation by experts (orthodontists), unvalidated audio and video quality assessment | Within-group comparisons | Care |
| Austin et al., 2010 | National Birth Defects Prevention Study (USA) | CL / CP / CLP | 2-7 years | Child with microtia or craniosynostosis | 253 mothers | Unvalidated survey completed by telephone | Within-group comparisons | Care |
| Bates, 2019 | Single centre (UK) | CL / CP / CLP | 10-16 years | Not reported | 23 participants:  5 children,  9 parents  (1 father, 5 mothers, 3 mother/father dyads), 9 professionals | Semi-structured interviews | N/A | Care |
| Bellon-Harn, 2024 | YouTube videos  (Global) | CL / CP / CLP | N/A | Duplicates, title or description did not  include the terms speech, resonance, hearing, feeding, videos consisting of promotional advertisement, PowerPoints lectures, talk shows, professional workshops, book reviews, and news channels, videos having parent or professional  testimonials without direct instructional content | 33 videos | Patient Education Material Assessment Tool-AudioVisual, DISCERN instrument, Video Power Index | None | Care |
| Bennett et al., 2018 | Single centre (USA) | CL / CP / CLP | 5-19 years | Non-English speaking | 60  (49 female, 11 male) | Barriers to Care questionnaire, Cleft Evaluation Profile | None | Care |
| Bennett et al., 2020 | Single centre (USA) | CL / CLP | 8+ years | (1) Non-English speaking, (2) under 8y, (3) isolated CP, (4) intellectual disability or (5) syndromic diagnosis. | 62  (31 patients aged 8+, 31 caregivers) | Semi-structured interviews | N/A | Care |
| Berger & Dalton, 2011 | Multi-centred (UK) | CL / CP / CLP | 11-16 years | Cognitive, language, or  communication difficulties | 191  (100 mothers, 91 adolescents) | Strengths and Difficulties Questionnaire (SDQ), General Well-being Scale (GWBS), KIDCOPE, Brief COPE, Satisfaction with Appearance Questionnaire (SWA), Childhood Experience Questionnaire (CEQ), Family Support Scale, Parenting Stress Index Life Stress subscale | None | Emotional |
| Berggren et al., 2012 | Single centre (Sweden) | CL, CLP | Not reported | Isolated CP | Prenatal group: 36 families  Postnatal group: 46 families | Unvalidated questionnaire | Within-group comparisons | Care |
| Breuning et al., 2021 | Single centre (Canada) | CL / CP / CLP | 8months – 6years | Non-English speaking, syndromic patients | 14 families  (3 w/ both parents and child, 11 mothers only) | Semi-structured interviews | None | Emotional  Social  Care |
| Brichacek & Matic, 2021 | Single centre (Canada) | CL / CLP | 0-6 years | Not reported | 37 parents  (non-specified mothers vs fathers) | Unvalidated open-ended survey | None | Emotional  Social  Care |
| Britton et al., 2011 | Single centre (UK) | CL / CP / CLP | 0-6 years | Not reported | 90 parents (non-specified mothers vs fathers) | Unvalidated questionnaire | None | Care |
| Cassell, 2012 | Multi-centred (USA) | CL / CP / CLP | 0-6 years | Child with OFC died, child was born or lived out of state, adopted children | 248 mothers | Unvalidated survey + questions extracted from Barriers to Care Questionnaire | None | Care |
| Cassell, 2013 | Multi-centred (USA) | CL / CP / CLP | 2-6 years | Child  with OFC died, child was born or lived out of state | 245 mothers | Unvalidated survey | None | Care |
| Cassell, 2014 | Multi-centred  (USA) | CL / CP / CLP | 2-6 years | Child with OFC died, child was born or lived out of state, adopted children | 245 mothers | Unvalidated survey | None | Emotional  Care |
| Collett et al., 2012 | Single centre  (USA) | CL / CP / CLP | 5-9 years | Did not speak English or Spanish, known syndrome or Mendelian-inherited disorder | 93 families with CL/P  124 controls | Child Behavior Checklist, PedsQL 4.0, Social Competence Scale, Parenting Stress Inventory | Control group | Emotional |
| Costa et al., 2019 | Charitable organisation  (UK) | CL / CP / CLP | Not reported | Not reported | 470 parents (92% mothers, 8% parents) | Unvalidated mixed-methods survey | None | Emotional  Care |
| Costa et al., 2020 | Multi-centre  (UK) | CL / CP / CLP | <1 year | Not reported | 517 parent dyads | PedsQL-HSGM | N/A | Care |
| Coste et al., 2022 | Single centre  (France) | CL / CP / CLP | 0-1 year | Not reported | 124 mothers (114 at 12-month follow-up) | Unvalidated survey (workshop evaluation) | Published normative data | Social  Care |
| Costa et al., 2023 | Online recruitment (UK) | CL / CP / CLP | Mean age (months): 5.30 | Child born with CL/P between January and June 2020 | 14 parents  (10 mothers, 4 fathers) | Semi-structured interviews | N/A | Emotional  Care |
| Crerand et al., 2015 | Multi-centred (USA) | CL / CP / CLP | 7.5-18 years | Inability to  read at a second-grade level, diagnosis with an  incomplete cleft lip without cleft of the alveolus, or  diagnosis of craniofacial syndrome  or other complex medical conditions | 1,200 parent-child dyads | Family Environment Scale (FES) | Published normative data | Emotional  Social |
| Cronin et al, 2021 | Non-clinical sample (Australia) | CL / CP / CLP | 2-3 years | Child not aged between 2–4 years | Expanded network of 7 children  (7 mothers, 6 fathers, 4 grandmothers, 2 grandfathers, 1 sibling, 1 aunt and 3 educators) | Ethnographic study  (semi-structured interview, case history interview, videos, recording of mealtimes, photos and field notes) | N/A | Emotional  Social  Care |
| Czajeczny et al., 2021 | Single centre (Poland) | CL / CP / CLP | Under 5 years | Not reported | 78  (69 women, 9 men) | Inventory for Measuring  Coping with Stress (Mini-COPE), the Family Resilience Assessment Scale (FRAS), and the Positive and Negative Affect Schedule (PANAS) | None | Emotional  Social |
| Dabit et al., 2024 | Multi-centred (USA) | CL / CP / CLP | 4-9 years | Additional major defects | 294 biological mothers | Mental Health Inventory, Aggravation in  Parenting Scale | Published general population data | Emotional |
| Davies et al., 2019 | Multi-centred (UK) | CP | 12-16 weeks | Infants with cleft lip +/- cleft palate, associated syndrome, breathing intervention required feeding intervention required, infants born prematurely, known cardiorespiratory disease, CL/P history in family | 27 parents | Semi-structured interviews | Within-group comparisons | Emotional  Care |
| Dean et al., 2019 | Single centre (USA) | CL | Not reported | Not reported | 94 caregivers  (8 males, 85 females) | Unvalidated survey | Within-group comparisons | Care |
| de Cuyper et al., 2019 | Single centre (Belgium) | CL / CP / CLP | 6 months – 6 years | Not reported | 45 families | Impact  on Family Scale (IOFS), Family Impact Scale (FIS), Care-Related Quality of Life Instrument (CarerQoL) | None | Emotional |
| de Pascalis et al., 2017 | Single centre  (UK) | CL / CP / CLP | 0-9 weeks | Not reported | Cleft: 30 mother-infant dyads  Control: 20 mother-infant dyads | Eye-tracking, General Areas of Interest (AOIs) & Facial AOIs | Control group | Emotional |
| Despars et al., 2011 | Single centre (Switzerland) | CL / CLP | 0-1 year | *Clinical group*: associated genetic syndrome, non-visibility of the cleft,  parental psychiatric illness, difficulty speaking French.  *Control group:* difficulties during  pregnancy or delivery, somatic abnormalities, parental psychiatric illness, difficulty  speaking French | 58 mothers  (22 cleft, 36 controls) | Working Model of the Child Interview (WMCI), The Impact of Event Scale (IES) | Control group | Emotional |
| Dissaux et al., 2021 | Multi-centred (France) | UCLP | 8-14 years | Isolated cleft lip or  palate, bilateral cleft syndrome,  associated malformation, psychological disorder, mental delay making them unable to read and/or understand | 56 families  (23A, 33B) | Unvalidated questionnaire | Within-group comparisons | Care |
| Douglas, 2012 | Multi-centred (UK) | CL / CP / CLP | Pregnancy | Not reported | 14 expectant parents | Unvalidated pilot-group workshops | N/A | Social |
| Esmonde et al., 2018 | Single centre (USA) | CL, CLP | 2-84 days at first clinic appointment | Patients not recommended  for NAM treatment by craniofacial team | Parents of 135 children  (77% males) | Adherence to NAM therapy | Within-group comparisons | Care |
| Feragen et al., 2017a | Multi-centred (Denmark, Finland, Sweden, Norway, UK) | UCLP | 5 years | Not reported | Parents of 356 children  (119 girls, 237 boys) | Cleft Evaluation Profile (CEP), unvalidated Scandcleft parent questionnaire | None | Emotional  Social |
| Feragen et al., 2017b | Multi-centred (Denmark, Finland, Sweden, Norway, UK) | UCPL | 5 years | Not reported | Parents of 356 children  (119 girls, 237 boys) | Unvalidated Scandcleft Questionnaire | None | Care |
| Forer et al., 2023 | Single centre (Israel) | CL, CP, CLP | 7-20 years | Syndromic cleft, not undergoing orthodontic treatment | 64  (63 parents, 1 orthodontist) | Cleft  Hearing, Appearance and Speech Questionnaire | Within-group comparisons | Care |
| Francisco et al, 2021 | Single centre (Portugal) | CL / CP / CLP | 8-27 years | Patients with cognitive disorders, craniofacial syndromes,  multiple dental loss, untreated dental caries, periodontal disease, severe facial trauma,  chronic pain, prior orthodontic treatment | 226 parents  (111 with cleft, 115 controls) | Oral Health Impact Profile-14 (OHIP-14), Family Impact Scale (FIS) | Control group | Emotional  Social |
| Gassling et al., 2014 | Single centre (Germany) | CL / CP / CLP | Mage = 9.00-10.6 | Not reported | 55 families (55 mothers, 55 fathers, 55 children) | Unvalidated analysis of the intra-familial interaction (video recording) | CLP vs healthy vs children with migraine | Emotional |
| Gibson et al., 2021 | Single centre (USA) | CL, CLP | Not reported | No palatal  or alveolar involvement | 106 patients | Patient’s records | N/A | Care |
| Gkantidis et al., 2013 | Single centre (Greece) | UCLP | Not reported | Patients with  syndromes, other congenital anomalies, psychological disorders | 12 young adults with UCLP and their parents, 24 laypersons, 6 orthodontists, 6 maxillofacial surgeons | Unvalidated questionnaire | General population | Care |
| Gkantidis et al., 2015 | Single centre (Greece) | CL, CP, CLP | 9-33 years | <9 years, syndrome, other congenital anomalies, learning difficulty | 33 patients and 30 parents | Unvalidated questionnaire | Within-group comparisons | Social  Care |
| Greives et al., 2017 | Social media websites  (Global) | CL / CP / CLP | Not reported | Not reported | 112 parents | Unvalidated survey | None | Emotional  Care |
| Grollemund et al., 2020 | Multi-centred (France) | CL / CP / CLP | 0-1 year | Isolated CP, child over 4 months at T0 | 158 infants and their parents | Alarm Distress Baby Scale (ADBB), Parenting Stress Index (PSI), Edinburgh Post-partum Depression Scale  (EPDS), The Impact on Family Scale (IOFS) | General population | Emotional |
| Habersaat et al., 2013 | Single centre (Switzerland) | CL, CLP | 2-12 months | Children with associated disorders or medical complications, cleft not visually apparent, parents who do not speak  French, parental history of psychiatric disorder | Cleft: 40 Controls: 45 | Interactive play coded using the Care Index, Perinatal Posttraumatic Stress Questionnaire (PPQ), the ‘‘strange situation’’ (SSP) | Control group | Emotional |
| Habersaat et al., 2014 | Multi-country (Benin + Switzerland) | CL, CP, CLP | Mean age in months  Swiss: 2.28  Benin: 34.9 | Children  with other malformations or medical complications | Benin: 36 mothers  Swiss: 40 mothers | Semi-structured interviews, Perinatal  Post-traumatic Stress Questionnaire, Beck Depression Inventory | Within-group comparisons | Emotional  Social |
| Habersaat et al., 2018 | Single centre (Switzerland) | CL, CLP | 2-60 months | Children with associated disorder or  medical complications, cleft not visually apparent, parents with a history of psychiatric disorder, not  sufficiently fluent in French | Cleft: 30  Controls: 14 | Working Model of the Child  Interview [WMCI], Parent  Development Interview [PDI], Parenting Style and Dimensions Questionnaire (PSDQ) | Control group | Emotional |
| Hansson et al., 2013 | Single centre (Sweden) | CL / CP / CLP | Not reported | Not reported | 33 parents | Unvalidated questionnaire | Published data from previous studies | Emotional  Social  Care |
| Hennocq et al., 2018 | Multi-centred (Paris, Nantes and Moscow) | CL, CLP | Age at  surgery was 6.722 months, not reported for 15 patients | Incomplete clefts, syndromic,  cognitive and/or motor impairment | 72 patients  (41 Paris, 21 Moscow, 10 Nantes) | Unvalidated questionnaire | None | Care |
| Hopkins et al., 2016 | Single centre (USA) | CL / CP / CLP | Not reported | Not reported | 12 parents  (8 mothers, 4 fathers) | Semi-structured interviews | N/A | Care |
| Huang et al., 2013 | Online support groups  (Global) | CL, CP, CLP | ≤1 year | Child over 1 year, syndromic cleft | 5 mothers | Unvalidated survey | None | Care |
| Jeong et al., 2013 | Single centre (North Korea) | CL / CP / CLP | 2 months – 17 years | Not reported | 36 mothers | Beck  Depression Inventory, Beck Anxiety Inventory, Parenting Stress  Inventory | General population | Emotional |
| Jodeh, 2019 | Single centre (USA) | CL, CP, CLP | ≤10 years | Not reported | 60 parents  (44 mothers, 16 fathers) | Unvalidated questionnaire | None | Care |
| Johns et al., 2018 | Single centre (USA) | CL / CP / CLP | 5.1 – 6.9 weeks | Additional medical concerns in the child | 206 mothers | Edinburgh Postnatal Depression Scale (EPDS) | General population | Emotional |
| Kaye et al., 2019 | Single centre (USA) | CL / CP / CLP | 1 – 2 years | Mothers who did not reside  with their infant during infancy | 50 mothers | Unvalidated telephone survey | None | Care |
| Kaye et al., 2022 | Single centre (USA) | CL, CP, CLP | < 18 years | Patients with incomplete records  related to their Child Protective Services (CPS) referral history | Parents of 25 patients with history of CPS referral | Patients’ records | N/A | Emotional |
| Ke et al., 2013 | Multi-centred (UK) | CL / CP / CLP | Not reported | Not reported | 16 parents | Semi-structured telephone interviews | N/A | Emotional  Care |
| Khouri et al., 2018 | Single centre (USA) | CL / CP / CLP | Not reported | Non-English speaking | 25  (22 mothers, 3 fathers) | Unvalidated survey completed by telephone | None | Social |
| Knapke et al., 2010 | Single centre (USA) | CL / CP / CLP | <1yr | Syndromes (excepting Stickler  syndrome or Van der Woude syndrome) | 17 parents (15 mothers, 2 fathers) | Unvalidated telephone interview | None | Care |
| Kramer et al., 2009 | Multi-centred (Germany) | CL / CP / CLP | 8-12 years | Syndromic OFC, receiving other medical care within last 3 months | 132 families  (81 male patients, 51 female patients) | Impact on family Scale (IOFS), KINDL Questionnaires | Age- and sex-matched control group | Emotional |
| Kuttenberger & Polska, 2010 | Single centre (Switzerland) | CL / CP / CLP | 10 weeks-19.5 years | Not reported | 73 parents | Unvalidated questionnaire | None | Emotional  Social  Care |
| Lentge et al., 2022 | Single centre (Germany) | CL / CP / CLP | 0-3 years | Not reported | 33 parents  (26 females, 7 males) | Parenting Stress Index, PSI + face-to-face interview | Published norm data | Emotional |
| Lindberg et al., 2024a | Single centre (Norway) | CL, CP, CLP | Not reported | Not able to speak, read, and write Norwegian, not referred by one month after birth | 70 families  (69 mothers and 57 fathers)  Intervention group (n =32)  Control group (n =38) | Parental Stress Index (PSI), Perceived Stress Scale (PSS-14), feeding questionnaire, survey of infant diets, weight percentiles | Control group | Emotional |
| Lindberg et al., 2024b | Single centre (Norway) | CL, CP, CLP | Not reported | Not able to speak, read, and write Norwegian, not referred by one month after birth | 70 families  (69 mothers and 57 fathers)  Intervention group (n =32)  Control group (n =38) | Use of Internet-Questionnaire, Quality of Discharge Teaching Scale (QDTS), Post Discharge Coping Difficulty  Scale (PDCDS), response on follow-up by health professionals | Control group | Care |
| Lindeberg & Berglund, 2014 | Single centre (Norway) | CL / CP/ CLP | Not reported | Not reported | 12 mothers | Semi-structured interview | None | Emotional  Social  Care |
| Losier et al., 2020 | Single centre (Canada) | CLP | 13-75 months | Children with other diagnosed developmental / mental health disorders, severe disabilities, intellectual disabilities | 84 adoptive parents  (76 mothers and 8 fathers)  29 children with CLP  55 controls | Unvalidated questionnaire, the Strange Situation protocol for children aged 12 to 24 months, the separation-reunion procedure adapted for children aged 24 to 72 months | Control group | Emotional |
| Maarse et al., 2018 | Single centre (Netherlands) | CL / CP / CLP | Not reported | Non-Dutch speaking, advanced gestation, not living in the Netherlands | 85 individuals  (45 mothers, 40 partners) | Unvalidated questionnaire | None | Emotional  Social  Care |
| Macho et al., 2017 | Single centre (Slovakia) | CL / CLP | 2 months - 1 year | Not reported | 40 families  (20 CL, 20 CLP) | Impact on Family Scale | None | Emotional  Social |
| Madhoun et al., 2020 | Variety of recruitment methods  (USA) | CL, CP, CLP | 8-14 months | Failure to answer initial screening questions, infant not within  defined age range, adopted, mother living outside the USA, English was not the primary language | 150 mothers | Unvalidated survey | Within-group comparisons | Social  Care |
| Madhoun et al., 2021 | Single centre (USA) | CL / CP / CLP | 1-12 weeks | CLP group: known genetic disorder or syndrome, except for  mild PRS.  Control group: genetic disorder or  syndrome, significant feeding or swallowing disorder, chronic medical condition. Both groups: non-oral feeding methods. | 60  (30 with CLP, 30 without CLP) | (1) Feeding/Swallowing Impact Survey (FS-IS), (2) Parenting Stress Index, Fourth  Edition, Short Form (PSI-4-SF), and (3) Edinburgh Postnatal Depression Scale | Control group | Emotional |
| Marcus et al., 2022 | Multi-centred (10 European countries) | CL | 0-10 years | Not living in Europe, child <10 years | 247 caregivers of children with cleft lip, spina bifida, CHD and/or Down syndrome | Unvalidated online survey in 9 languages | Within-group comparisons | Care |
| Martin & Greatrex-White 2014 | Multi-centred (UK) | CP, CLP | Not reported | Babies  diagnosed with syndromes or respiratory problems | 50 mothers | Feeding diaries, Edinburgh Postnatal Depression Score (EPDS), unvalidated visual analogue scales | Within-group comparisons | Emotional |
| Martin et al., 2020 | Multi-centred  (UK) | CL, CP, CLP | Not reported | Not reported | Parents of 38 babies | Unvalidated questionnaire | Within-group comparisons | Care |
| McCorkell et al., 2012 | Single centre (UK) | CL / CP / CLP | 12 months – 8 years | Not reported | 20 parents  (16 mothers, 1 father, 1 grandmother) | Semi-structured interviews | N/A | Emotional  Care |
| McWilliams et al., 2022 | Variety of recruitment methods  (UK) | CL, CP, CLP | Not reported | Staff who were currently or who had recently been on temporary enforced leave | 27 healthcare providers and charity staff | Semi-structured interviews | N/A | Care |
| Montirosso et al., 2012 | Single centre (Italy) | CL / CLP | 2 months | Prematurity,  other syndromes,  mothers with learning difficulty, psychiatric disorder or addiction, teenage parent, single parent, non-Italian nationality | Mothers of 25 infants  (21 males, 4 females)  25 controls | Family SES, Infant  Behavior Questionnaire-Revised (IBQ-R), Beck Depression Inventory (BDI), Global Rating Scales of Mother–Infant Interaction (GRS) | 25 age-matched healthy infants | Emotional |
| Murray et al., 2018 | Recruitment methods unclear  (UK) | CL, CLP | Not reported | Not reported | Parents of 10 children  Control group = 20 | Videotaped interactions, eye tracking, Edinburgh Postnatal Depression Scale (EPDS) | Control group | Emotional |
| Nelson & Kirk, 2013 | Single centre (UK) | CL / CP / CLP | 20 weeks – 21 years | Non-English speakers, families with challenging circumstances | 35 parents  (24 mothers, 11 fathers) | Semi-structured interviews | N/A | Care |
| Nelson et al., 2012a | Single centre (UK) | CL / CP / CLP | 20 weeks – 21 years | Non-English speakers, families with challenging circumstances | 35 parents  (24 mothers, 11 fathers) | Semi-structured interviews | N/A | Emotional  Care |
| Nelson et al., 2012b | Single centre (UK) | CL / CP / CLP | 20 weeks – 21 years | Non-English speakers, families with challenging circumstances | 35 parents  (24 mothers, 11 fathers) | Semi-structured interviews | N/A | Emotional  Social  Care |
| Nes et al., 2014 | Multi-centred (Norway) | CL / CP / CLP | 0-3 years | Pregnancies not ending in live births, children with severe congenital anomalies, missing responses on all relevant variables | 179 mothers | (1) Satisfaction With Life Scale (SWLS), (2) short version of the 25-item Hopkins Symptom Checklist (SCL-25) | Within-group comparisons | Emotional |
| Nidey et al., 2015 | Multi-centred (USA) | CL, CP, CLP | 0-17 years | No history of genetic conditions, parents of multiple affected  children | 287 parents  (171 mothers and 116 fathers) | Social Avoidance and Distress (SAD) scale, Fear of Negative Evaluation (FNE) scale, Rosenberg Self-Esteem (RSE),  Interpersonal Support Evaluation List (ISEL) | None | Emotional  Care |
| Niinomi et al., 2022 | Single centre (Japan) | CL / CP / CLP | 0-12 years | Syndromic CL/P, other  chronic or congenital diseases | 171 parents | Unvalidated survey | None | Emotional |
| Omiya & Yamazaki 2017 | Single centre (Japan) | CL, CP, CLP | Mean age = 18.7 years | Questionnaires answered by the father or other relatives | 293 mothers | Unvalidated questionnaire, Perceived Positive Change (PPC) scale, Subjective Social Capital (SC) scale, Japanese version of the SOC-13 | None | Emotional |
| Ranganathan et al., 2019 | Single centre (USA) | CL, CP, CLP | 5-19 years | Not able to read and respond independently in English | Children – 100  Caregivers – 100  Surgeons – 10  Control observers - 10 | Cleft Evaluation  Profile (CEP), unvalidated questionnaire, photographs eliciting rating, unvalidated questionnaire to surgeons and control | Within-group comparisons | Care |
| Robbins et al., 2010 | Multi-centred (USA) | CL, CP, CLP | 2-7 years | Child not living  with biological mother, family moved out of state, diagnosis of other craniofacial condition | 235 mothers | Unvalidated survey | Within-group comparisons | Emotional  Care |
| Roth et al., 2021 | Multi-centred (Taiwan and Germany) | UCLP | Not reported | Not reported | Parents of 117 children  Germany: 15 mothers, 13 fathers  Taiwan: 38 mothers, 34 fathers | Unvalidated questionnaire | Within-group comparisons | Care |
| Sato et al., 2021 | Multi-centred (Japan) | CL, CP, CLP | Not reported | Infant sex not recorded, non-classified orofacial clefts, other congenital anomalies or syndromes, mothers with a history of depression or antipsychotic drug use in the past year, participants who did not answer dependent-variable questions more than 3 times | 148 mothers of infants with CL/P  84,454 control group | Kessler Psychological Distress  Scale, Edinburgh Postnatal Depression Scale | Control group | Emotional |
| Scheller et al., 2020a | Single centre (Germany) | CL / CLP | 9 months – 27 years | Syndromic cleft, other malformations or chromosomal aberration | 84 mothers | Unvalidated survey | None | Emotional  Social  Care |
| Scheller et al., 2020b | Single centre (Germany) | CL, CP, CLP | 0-1 year | Children with a syndromic cleft or other malformations | 84 mothers | Validated measures, not reported | None | Emotional  Care |
| Searle et al., 2016 | Multi-centred (UK) | CL / CP / CLP | Not reported | Not reported | 24 families  (15 mothers, 2 fathers, 7 couples) | Semi-structured interviews | N/A | Emotional  Social  Care |
| Searle et al., 2018 | Multi-centred (UK) | CL / CP / CLP | 7 months – 19 years | Not reported | 25 families  (16 mothers, 3 fathers, 12 couples) | Semi-structured interviews | N/A | Care |
| Sell et al., 2023 | Multi-centred (Ireland and UK) | CP, CLP | 3-7 years | Not reported | 21 parents  (3 fathers, 17 mothers) | Focus group, semi-structured interviews | N/A | Care |
| Shipe et al., 2016 | Single centre (USA) | CL / CP / CLP | Mean age at adoption= 2.3 (1.8) | Had primary surgery elsewhere | 20 caregivers  (11 female, 9 male) | Semi-structured interviews | N/A | Emotional  Care |
| Shuttlewood et al., 2014 | Multiple recruitment methods  (UK) | CL, CP, CLP | 1-23 years | Insufficient understanding of English, known  significant mental health problems,  known additional physical health problems, child cared for by persons outside  biological extended family | 179 parents (74.9% mothers) | Parental  Appraisal of Cleft Questionnaire, Psychological General Well-Being Index (PGWBI), Socially Desirable  Response Set (SDRS-5), Social Support  Questionnaire (SSQ) | None | Emotional  Social  Care |
| Sischo et al., 2015 | Multi-centred (USA) | CL / CLP | 0-1 year | Non-English or non-Spanish-speaking, syndrome, other major medical issues, caregivers with major psychiatric disorder | 68 caregivers  (62 mothers, 6 fathers) | Semi-structured interviews | N/A | Emotional  Social  Care |
| Sischo et al., 2016 | Multi-centred (USA) | CL / CLP | 0-1 year | Non-English or non-Spanish-speaking, syndrome, other major medical issues, caregivers with major psychiatric disorder | 118 caregivers  (107 females, 11 males) | (1) Semi-structured interviews, (2) The Patient Health Questionnaire (PHQ-9), (3) Generalized Anxiety and Depressive symptom scales (GAD-7), (4) The Parenting  Stress Index—Short Form (PSI–SF), (5) LOT-R, (6) The Family Environment  Scale (FES), (7) Coping  Health Inventory for Parents (CHIP) | Within-group comparisons | Emotional  Social  Care |
| Snyder & Rushcello, 2019 | Single centre (USA) | CL / CP / CLP | 6 months – 7.5 years | Diagnosis of syndrome or sequence | 26 families  (20 mothers, 1 father, 5 couples) | Unvalidated 29-items questionnaire | None | Social  Care |
| Southby et al., 2022 | Multi-centre (UK) | CP, CLP | 5.7 months–12.4 years | Not reported | 212  (153 mothers, 59 fathers/other caregiver) | Unvalidated questionnaire | None | Care |
| Spoyalo et al., 2021 | Single centre (USA) | CLP | 0-11 years | Syndromic CLP | 24 parents  (1 couple, 19 mothers, 3 fathers) | Focus group and telephone interviews | N/A | Care |
| Srivastav et al., 2022 | YouTube videos  (Global) | CL, CP, CLP | Not reported | Feeding videos for children without CL/P, feeding videos for children with other healthcare needs, videos providing  information not about infancy | 42 videos | Unvalidated protocol | N/A | Care |
| Stock & Rumsey, 2015 | Variety of recruitment methods  (UK) | CL / CP / CLP | 4.5 months – 24 years | Not reported | 15 fathers | Semi-structured interviews | N/A | Emotional  Social  Care |
| Stock et al., 2018 | Online support groups  (UK) | CL, CP, CLP | Not reported | N/A | 150 parents/ caregivers  64 adults with CL/P  Online survey: 39 parents/ caregivers, 22 adults with CL/P | Content analysis using screen capture program  Unvalidated online survey | N/A | Emotional  Social  Care |
| Stock et al., 2019a | Charitable organisation  (UK) | CL / CLP | Not reported | Not received antenatal diagnosis of CL/CLP | 574 parents  (88% mothers, 12% fathers) | Unvalidated online survey | None | Emotional  Care |
| Stock et al., 2019b | Charitable organisation  (UK) | CL / CLP | Not reported | Not received antenatal diagnosis of CL/CLP | 217 parents (90% mothers, 10% fathers) | Unvalidated online survey | None | Emotional  Care |
| Stock et al., 2020a | Multi-centred  (UK) | CL / CP / CLP | <1 year | Not reported | 1163 parents (644 mothers, 519 fathers) | Outcomes: PedsQL-FIM, PSS, HADS and CEN-Q  Predictors: PedsQL-HSGM, LOT-R, SRRS, RSS | Published normative data | Emotional  Social |
| Stock et al., 2020b | Single centre (UK) | CL, CP, CLP | Not reported | Not reported | Focus group (n = 4):  14 caregivers  10 young people  7 adults  4 healthcare professionals  Feedback form: 924  522 caregivers  247 children and young adults  44 adults  111 volunteers  Online survey: 82 stakeholders | Focus groups, unvalidated surveys | N/A | Social |
| Stone et al., 2010 | Multi-centred (USA) | CL / CP / CLP | 0-18 years | Not reported | 20 parents  (15 mothers, 5 fathers) | Focus groups | N/A | Emotional  Social  Care |
| Thompson et al., 2021 | Multi-centred (New Zealand) | CL / CP / CLP | 5-12 years | Known syndrome | 397 | QoL subscale of PedsQL | Within-group comparisons | Emotional |
| Tierney et al., 2013 | Multi-centred (UK) | CP / CLP | 0-11 years | Syndrome, no OME, non-English speaking, significant psychosocial difficulties | 37 families  (6 couples, 30 mothers, 1 father, 37 children - 16 females, 21 males) | Semi-structured interviews | N/A | Care |
| Tierney et al., 2015a | Multi-centred (UK) | CP / CLP | 0-11 years | Syndrome, no OME, non-English speaking, significant psychosocial difficulties | 37 families  (6 couples, 30 mothers, 1 father, 37 children - 16 females, 21 males) | Semi-structured interviews | N/A | Emotional  Care |
| Tierney et al., 2015b | Variety of recruitment methods  (UK) | CP | 0-4 years | Non-English speakers | Parents of 17 children  (1 couple, 16 mothers) and 3 specialist nurses | Semi-structured interviews | N/A |  |
| Tsuchiya et al., 2019 | Multi-centred (Japan) | CL / CP / CLP | 1 year | Other congenital disease | 79,140 mother-child dyads | The Mother-to-Infant Bonding  Scale, Kessler Distress Scale (K6) | Published normative data | Emotional |
| Ueki et al., 2019 | Single centre (Japan) | CL / CP / CLP | 0-12 years | Other chronic disease or syndrome | 64 couples | Scale to Measure Resilience in Child Care (SMRCC), unvalidated questionnaire | Within-group comparisons | Emotional  Care |
| Van Dalen et al., 2021 | Single centre (Netherlands) | CL / CLP | 0-12 years | Isolated cleft palate or cleft  alveolus, non-sufficient knowledge of the Dutch  language, children >12 years | 309 parents  (173 mothers, 136 fathers) | (1) NOSI-K – Dutch shortened version of the Parenting Stress Index, (2) corresponding depression anxiety and hostility subscales of the Dutch  Symptom Checklist – 90 (SCL-90), (3) the Child Behavior Checklist: ages  1.5 to 5 (CBCL 1.5-5), (4) the  Child Behavior Checklist: ages 6 to 18 (CBCL 6-18) | Published normative data | Emotional |
| Van Lierde et al., 2012 | Single centre  (Belgium) | UCLP | 10-17 years | Syndromic UCLP, secondary pharyngeal surgery, cognitive deficiency, neuromotor dysfunction or residual hard palate fistula and specific hearing threshold | Parents of 43 children with UCLP (28 boys, 15 girls)  Controls: 43 families | Cleft Evaluation Profile (CEP) | Age and gender matched control group | Care |
| Wallace & Mattner, 2017 | Multi-centred (Australia) | CL / CP / CLP | 25-38 years | Child with multiple birth anomalies | 5 parents  (4 mothers, 1 father) | Semi-structured interviews | N/A | Emotional  Social  Care |
| Wogden et al., 2019 | Charitable organisation  (UK) | CL / CP / CLP | 12-25 years | Not reported | 30 participants: 11 young people (3 male, 8 female), 17 parents (13 mothers, 4 fathers),  5 professionals | Semi-structured interviews (young people) and unvalidated online survey (parents and HCPs) | None | Care |
| Zeytinoğlu et al., 2016 | Single centre (USA) | CL / CP / CLP | 1-4 years | Not reported | 17 fathers | Semi-structured interviews | N/A | Emotional  Care |
| Zeytinoğlu et al., 2017 | Single centre (USA) | CL / CP / CLP | 1-4 years | Inability to read and/or understand English, presence of a cognitive or physical disability, child diagnosed with other significant health problems | 17 couples | Semi-structured interview and the Revised Dyadic Adjustment Scale (RDAS) | None | Emotional  Social  Care |

**Table 2 –** Predictors of Parental Adjustment

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| **Risk Factors for Psychological Distress** | **Protective Factors for Psychological Distress** |
| A postnatal diagnosis  Delayed diagnosis  Older age at time of diagnosis  Pre-existing mental health condition  Lower annual income  Fewer years of parental education  Minority ethnic group  Lack of understanding about aetiology  Self-blame  Difficult interactions with health professionals  A negative appraisal of CL/P  Lack of relevant and timely information  Perceived stigma  Feeding difficulties  Presence of a syndrome  Avoidant/restraint coping  Burden of care  Poor care coordination  Childcare difficulties  Type of health insurance  Surgical complications  Hearing difficulties  Low speech intelligibility  Behavioural problems in the child  Intensive treatment stage  Lack of shared decision-making | Positive outlook on life  Cleft lip only  A healthy parent-infant bond  Problem-based coping  Receiving support from friends and family  Engaging in peer support  Relationship satisfaction  Meaning making  Being under the care of a specialist team  Taking an active role in treatment  Measurable treatment progress  Healthcare satisfaction |

**Table 3:** Recommendations for Clinical Practice and Future Research

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| **Recommendations for clinical practice** |
| Increased awareness of CL/P informational resources and referral pathways among non-specialist health professionals |
| Improved quality, relevance and accessibility of CL/P information for parents |
| Access to specialist feeding advice and equipment |
| Involvement of the father in healthcare appointments and decisions |
| Facilitation of effective support networks |
| Regular audit of services in collaboration with parents and families to identify areas of strength and opportunities for improvement |
| Routine psychological screening of known risk factors to identify parents and families in need of support |
| Evidence-based psychoeducational resources for parents to address common concerns at key stages in the treatment journey |
| Integrated specialist psychological support from diagnosis onward |
| **Recommendations for future research** |
| An assessment of what information different families need and at which stage |
| Inclusion of families from diverse ethnic and socioeconomic backgrounds |
| Further examination of the factors that contribute to personal growth |
| Additional research into effective shared decision-making |
| Multicentre, prospective, longitudinal studies with sufficient sample sizes and appropriate control/reference groups |
| Application of relevant theories and models |
| Development and evaluation of evidence-based psychological interventions |
| Cross-condition learning which draws on sources outside the immediate field |