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 families[1-3] (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 unit[1,4-5] (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 needs[6].

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 globally[7] (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 required[8] (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/P[9] (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 conditions[9]. 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 al[9]. 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).

1.1. 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 future[11-18]. Some parents also expressed delight[13], while others felt the joy of having a baby had been somewhat diminished by the diagnosis[11,19]. 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 addressed[15,20-22], many grappled significantly with definitions of normality, perfection and difference[13,18]. 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 happiness[20]. 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 diagnosis[12,23], others identified greater concerns in those receiving a diagnosis postnatally[15,17,24]. Parents' acceptance of their child's appearance was also more negatively impacted if they received the diagnosis after birth[17]. In contrast, receiving a prenatal diagnosis of CL/P gave parents more time to adjust and prepare[11,15,18,25]. Detection rates of cleft palate only were more variable, with some reports of delayed diagnosis resulting in parental distress[15,26-27]. A lack of understanding of the aetiology of CL/P was associated with greater self-blame in parents[12], which in turn predicted poorer general well-being, depression, elevated stress and anxiety scores, and a negative impact on parent-infant bonding[28-31].

1.2. 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 differences[9,11,15,17-18,22,32-37]. 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/P[11-12,14-15,18-19,21-22,25,36,38-41].

1.3. Feeding Difficulties

Feeding difficulties were the cause of frustration, anxiety, distress, sadness and despair for parents[14,26,42]. Some mothers also described a sense of loss at not being able to breastfeed their child as they had planned[18]. Feeding difficulties were described by mothers as traumatic in one study[41], while in two others, parents reported feeling personally responsible for their child's failure to thrive[27,43]. Using breast pumps was considered to be stressful and time-consuming[14]. Problems with feeding and a longer duration of feeds predicted poorer maternal health-related quality of life in two studies[43-44].

1.4. 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 without[45-46]. In contrast, three other studies identified significantly more disengaged attachment representations and negative interaction patterns exhibited by mothers of children with CL/P[47-49]. One study found that mothers of infants with cleft lip gazed at their infants' faces less often than controls[50]. Mothers also reported bonding insecurities, concerns about a lack of physical closeness and reduced maternal instinct[14,41]. 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 initiative[51]. Parents qualitatively reported no impact and/or a positive impact on bonding in one study exploring parents' early experiences[52]. Nasoalveolar Moulding (NAM) treatment was found to both positively and negatively impact bonding according to parent report[53].

1.5. 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 al[54]., 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 communicate[54]. Berger and Dalton found hearing difficulties to account for 6.6% of the variance in parental wellbeing[28]. In two other qualitative studies, parents expressed frustration at their child not being able to communicate intelligibly with others[15,41]. Speech was a particular challenge for parents of internationally adopted children, given the language barrier[33,55]. 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 depression[56-59].

1.6. 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 life[23,48,58-64]. In contrast, other studies identified elevated stress, anxiety, and depression, alongside more symptoms of emotional difficulties, post-traumatic stress and decreased quality of life[23,35,47,63,65-70]. Three studies identified elevated postnatal depression scores in mothers[24,43,71], with 11.7 percent of mothers reporting scores above the clinical cutoff in one study[24]. Mothers in this sample also reported feeling anxious, scared and sad, with 1.5% endorsing minor intent for self-harm[24]. Despite elevated anxiety and depression scores compared to a normative sample, scores remained in the normal range in two studies[66,71]. Parents qualitatively described elevated levels of anxiety during the Covid-19 pandemic[72]. Other studies reported lower levels of stress, less anxiety, and better overall quality of life than control/reference groups[35,59,66]. Stress scores

fluctuated according to treatment stage in one study[35]. 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 work[24,28,59,62,66,73-74]. A positive life orientation was identified to protect parents from psychological distress[66]. Five studies found no variations in parental well-being according to cleft type[23,37,59,66,75], while others identified a higher impact of cleft lip and palate compared to cleft lip or cleft palate only[37,65,68,73,76], or a reduced impact for those with cleft lip only[58,77]. Mothers scored significantly less favourably than fathers on measures of mental health in some studies[66,73]. Psychological distress was found to reduce over time in two studies[63,77]. Parents in two studies specifically identified a need for dedicated emotional support[12,13].

1.7. 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 advocacy[14,18,35,53,78]. Active coping and seeking emotional and social support were associated with family resiliency in one study[31]. Coping strategies associated with lower resiliency and lower positive affect included restraint coping, substance use, self-blame and denial[31]. 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 relationships[15,22,29,53,79-80].

2. 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).

2.1. 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 professionals[13,22,33,36,52]. Curiosity from strangers and comments about their child's appearance were often experienced by parents as hurtful or distressing and could have a lasting impact[13,22,36,52,57]. 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 themselves[18,36]. Parents chose not to share photographs of their child prior to lip surgery in one study[18]. Some comments were perceived by parents to be more neutral or positive in nature[17-18,36], although well-intended comments could also be seen as trivialising[36]. Whispers, stares and pity were also reported[36]. 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 comments[36].

2.2. Marital Relationship

One study by Maarse and colleagues[20] 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 lessened[81]. Stock et al. also found relationship satisfaction to be protective against psychological distress for both mothers and fathers[66]. 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 cases[16,19]. Yet, if the couple were able to work together in times of stress, the marital relationship could grow in strength[14,16,1,9,41].

2.3. 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 conflict[65,67,82]. A total of 36.5% reported 'a lot' or 'some' impact of CL/P on family life in a study by Agnew and colleagues[75]. However, in two multicentre studies with >1,000 participants, scores of family cohesion, expressiveness, conflict were in the normal range[83] and both mothers and fathers reported more favourable scores for daily activities and family relationships compared to normative data[66]. Sischo and colleagues also identified greater family expressiveness and less conflict in families with CL/P compared to published norms[35]. 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 satisfaction[35,66,83].

2.4. Social Support

Several papers noted the importance of family support for parental adjustment to CL/P. This support could be emotional or practical in nature[14,36,44,57]. Utilising family support was seen as a core coping strategy in some studies[18,11,16,35,41], while unsupportive comments or behaviours by family members were harmful to parental well-being[22,36]. Similarly, changes in friendships could be hurtful for parents[36], while close friendships were found to be protective against depression in mothers[66]. 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 challenges[11,14,18,22,35,53,84-86]. Some non-profit organisations also provided physical resources such as feeding bottles and information leaflets, which parents found helpful[16]. In other cases, parents identified a lack of social support

and wanted information about and access to support organisations and local parent groups[13,38,87]. A large proportion of parents had also used the internet, including social media to access information about CL/P[22,40,88]. One study found a perceived lack of social support to predict depression, anxiety, less self-control and poorer well-being and vitality[29], while another found effective social support to be correlated with resiliency[31].

3. 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).

3.1. 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 family[15,17-18,20-22,25,27,38,41]. Many parents receiving a diagnosis antenatally had been asked to consider terminating the pregnancy without an opportunity to fully understand the diagnosis[20,22,26,89], while others had felt pressured to undergo amniocentesis with a view to terminating the pregnancy if a positive result was identified[11,89]. Those parents that came close to ending their pregnancy reported significant distress[20,89]. 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 teams[26-27]. A number of papers also described unhelpful, insensitive or dismissive reactions from non-specialist health professionals that had a lasting impact on parental well-being[15-16,18,21-22,26-27,41,89]. 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 inadequate[33]. In response to a lack of sufficient information, parents often utilised the internet and social media to learn more about CL/P and its treatment[11,13,16,21-22,27,38], yet the quality of online information was found to be highly variable[17,18,39-40,88,90-99]. Parents wanted reassurance from health professionals, counselling opportunities and consistent and accurate information to reduce their anxiety[11-12,20,25,38]. Families who were followed up early by a specialist nurse were less likely to utilise online support in one study[64]. Differences in the desired level and timing of information were observed[11,34,38,100], with some parents preferring not to view pre- and post-surgery photographs[16,38-39]. On the whole, parents felt reassured and much more informed once under the care of a specialist health professional or team[22,38,93,101].

3.2. 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 unprepared[11,14,87]. Non-specialist health professionals were also perceived to lack the expertise necessary to meet the child's feeding needs[14,41,102]. A large proportion of parents received no encouragement to breastfeed or were actively discouraged according to two studies[74,87], and rates of continuation fell behind the national average[44,74]. 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 discontinuation[74]. Yet, when parents were provided with specialist information, counselling, individualised lactation support and practical guidance, rates of continued breastfeeding were high[12,14,86,97]. 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 success[87].

3.3. 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 available[32,103]. The pursuit of treatment was also driven by a need to reduce the likelihood of real or anticipated social stigma[13,15,32]. Yet, parents also reported conflicting feelings about sanctioning treatment, particularly if the primary goal was to 'normalise' their child's appearance[18,22,32,35]. Parents worried about the risks of surgery, pain management and the physical and emotional impact of treatment on their child[11,18,35,55,104-105]. Trust in the medical team was therefore essential, and many parents chose to follow health professionals' treatment recommendations[11,13,15,19,22,32]. 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 option[16,106-107]. One study reported that parents wanted to be involved in decisions and to take a proactive role in their child's treatment[108]. 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 older[32,103,109].

3.4. 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 travelled[11,18,22,34,56,104-105,108,110-114]. This burden was greatest for parents with less education, parents belonging to an ethnic minority group and parents speaking languages other than English[56,111,115]. A greater impact of treatment predicted poorer global well-being, vitality and general health, and greater anxiety in one study[29]. 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 uncertainty[13,22,41,53]. Having continuous access to a highly experienced and specialist team alleviated some concerns and produced better parent-reported

outcomes[11,18,22,26,34,102,104,108,116-117], yet team intervention was still described by some parents as overwhelming[22,80,102]. 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 repeated[13,19,22,54,106,118-119]. Some parents stated they had not been prepared for the change in their child's appearance after surgery and had found this distressing[11,22]. 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 professionals[41,54,107]. 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 process[35,53,91]. 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 coverage[120-121]. Parents who did not complete NAM were less satisfied with the outcome of surgery[121]. Parents described needing to see measurable progress to support the efficacy of treatments and to make the burden of treatment worthwhile[41,80,104,106,108].

3.5. Healthcare Satisfaction

Overall, parents reported a high level of satisfaction with the care they and their child had received from the specialist CL/P team[22,33-34,39,56,64,102,104-105,100-111,117,122-123]. 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 development[25,104,124]. 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 professionals[25,104,124]. Fathers were found to be particularly dissatisfied with their access to credible information and their inclusion in medical appointments and support networks in four studies[22,64,73,123], which worsened during the Covid-19 pandemic[72]. During the pandemic, reduced contact with health professionals, changes to surgical protocols and surgical delays negatively impacted parents' experiences of care[72,125]. However, access to telehealth was broadly viewed as a potentially useful adjunct for future in-person care[72,125,126]. Eight studies specifically described parental satisfaction with the aesthetic and functional outcomes of NAM[35,53,102,127] and surgery[11,15,52,81]. 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 well[42,78,81-82,115,128-130]. Parents were less satisfied with treatment if the child had combined cleft lip and palate[82,111,115]. Parents and children were prone to disagreement, with parents being less satisfied with treatment outcomes and having a greater desire to pursue further surgery[81,128-131].

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 2009[9]. 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 review[9].

As identified in Nelson et al.'s 2012 review[9], 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 types[132]. 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 review[9], 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 adjustment[9], 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 review[9], 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 intervention[133], 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 tool[134]. 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 support[1]. 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 recommendations[9], 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 disability[135-138], 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 measures[139-140]. 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 field[133,141]. 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 conditions[1-5], 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

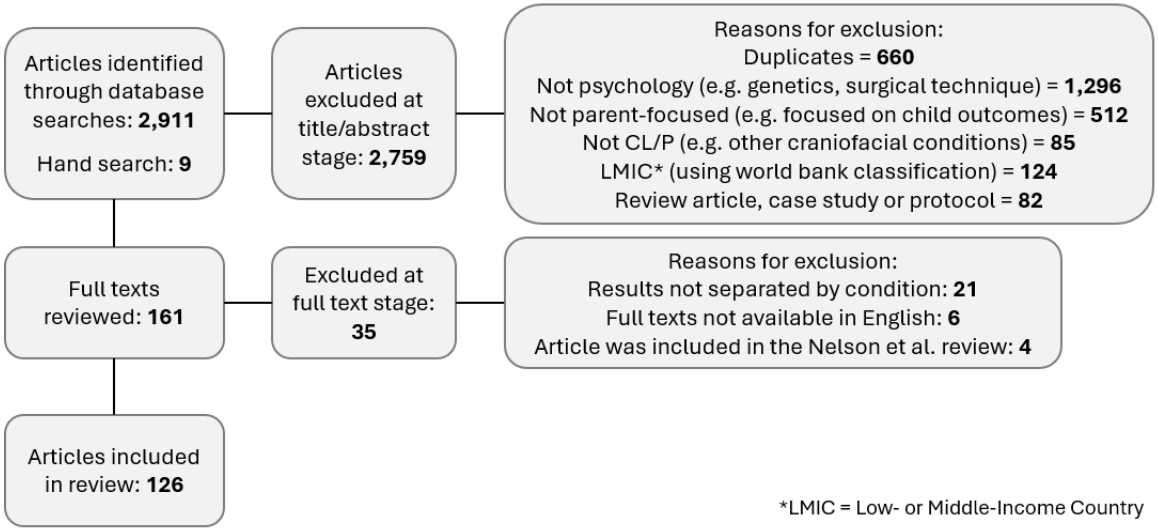

Table 1: Data Extraction Table

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	40 parents	Semi-structured interview	Within-group comparisons	Care

				additional procedures; inability to provide informed consent				
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	245 mothers	Unvalidated survey	None	Care

				with OFC died, child was born or lived out of state				
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	None	Emotional Social

						Resilience Assessment Scale (FRAS), and the Positive and Negative Affect Schedule (PANAS)		
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,	58 mothers (22 cleft, 36 controls)	Working Model of the Child Interview (WMCI), The Impact of Event Scale (IES)	Control group	Emotional

				parental psychiatric illness, difficulty speaking French				
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	Cleft: 30 Controls: 14	Working Model of the Child Interview [WMCI], Parent	Control group	Emotional

				medical complications, cleft not visually apparent, parents with a history of psychiatric disorder, not sufficiently fluent in French		Development Interview [PDI], Parenting Style and Dimensions Questionnaire (PSDQ)		
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)	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

					Control group (n =38)			
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	Unvalidated questionnaire	Within-group comparisons	Care

					Taiwan: 38 mothers, 34 fathers			
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	179 parents (74.9% mothers)	Parental Appraisal of Cleft Questionnaire, Psychological General Well-Being Index (PGWBI), Socially Desirable	None	Emotional Social Care

				cared for by persons outside biological extended family		Response Set (SDRS-5), Social Support Questionnaire (SSQ)		
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	Focus groups	N/A	Emotional Social

					(15 mothers, 5 fathers)			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

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

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