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Psychological Adjustment to Cleft Lip and/or Palate: A Narrative Review of the Literature

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Abstract

Objective: Adjustment to cleft lip and/or palate (CL/P) is multifaceted, involving several domains of psychological and social functioning. A substantial increase in research in this area has been evident in recent years, along with a preliminary shift in how adjustment to CL/P is conceptualised and measured. An updated and comprehensive review of the literature is needed in light of the rapidly expanding and changing field.

Design: A narrative review of 148 quantitative and qualitative studies published between January 2004 - July 2015.

Main Outcome Measures: Findings are presented according to five key domains of adjustment: Developmental Trajectory, Behaviour, Emotional Wellbeing, Social Experiences and Satisfaction with Appearance and Treatment. Data pertaining to General Psychological Wellbeing were also examined.

Results: The overall impact of CL/P on psychological adjustment appears to be low. Nonetheless, the review demonstrates the complexity of findings both within and across domains, and highlights recurring methodological challenges.

Conclusions: Research findings from the last decade are considered to be largely inconclusive, although some areas of emerging consensus and improvements in the approaches used were identified. Efforts to collect data from large, representative and longitudinal samples, which are comparable across studies and encompassing of the patient perspective, should be doubled.

Keywords: cleft lip and palate, development, behaviour, emotional, social, treatment, quality of life

Word count: approximately 12,597

Running title: Psychological adjustment to CL/P: A narrative review

Introduction

A cleft of the lip and/or the palate (CL/P) is one of the most common congenital conditions found in humans. Although the prevalence rate of CL/P varies substantially across different geographical areas and ethnic groups, it occurs in approximately one in every 500-700 births per year globally (World Health Organization, 2012). Although the most visible features of the cleft are normally repaired during the child's first year of life, facial scarring, functional difficulties and/or speech problems may remain. Throughout childhood, individuals born with CL/P will thus engage in a multidisciplinary treatment pathway, including plastic and maxillofacial surgery, otorhinolaryngology, orthodontics, speech and language therapy and psychology.

CL/P and its treatment can pose many challenges for those affected and their families, impacting upon several areas of psychological and social functioning. For individuals born with CL/P, ongoing treatment and medical appointments may represent a significant burden, in addition to the comments, questions, staring and teasing which may accompany a visible and/or audible difference (Rumsey & Harcourt, 2005). Previous research has thus highlighted potential difficulties for those born with CL/P in relation to social, emotional, cognitive and behavioural functioning (see Hunt, Burden, Hepper & Johnston, 2005 for the most comprehensive recent review). Tentative evidence for the impact of a number of interposing variables on individual adjustment, such as age, cleft type, gender, ethnicity, socio-economic status and the presence of other conditions additional to the cleft has also been presented (Hunt et al., 2005). Unfortunately, findings are contradictory and most existing reviews report significant methodological limitations which prevent firm conclusions from being drawn.

In spite of the number of review articles published during the last decade (Hunt et al., 2005; Collett & Speltz, 2007; Yazdy, Honein, Rasmussen & Frias, 2007; de Sousa, Devare & Ghanshani, 2009; Richman, McCoy, Conrad & Nopoulos, 2012; Klassen et al., 2012; Pavia & Andre, 2012; Zeytinoglu & Davey, 2012; Antonarakis, Patel & Tompson, 2013; Dimberg, Arnrup & Bondemark, 2015; Liddle, Baker, Smith & Thompson, 2015; Queiroz Herkrath, Herkrath, Rebelo & Vettore, 2015; Zhu, Jayaraman & Khambay, 2015), none have offered a comprehensive update in relation to psychological adjustment to CL/P since the overview provided by Hunt et al. in 2005. Of the reviews published since 2005, the majority have made repeated reference to literature which could be considered out of date, despite the high volume

of research which has been published more recently. Where this more recent literature has been considered, much of it has been excluded for not meeting stringent inclusion criteria, leaving only a small percentage of the total information published available for scrutiny. Given the well-known commonality of methodological challenges in this field, an inclusive approach to reviewing the literature may yield a better understanding of the work being carried out and of where the difficulties lie. In addition, several of these reviews have limited their focus to specific aspects of adjustment, such as neuropsychological, behavioural and academic functioning (Richman et al., 2012), aesthetic and psychosocial outcomes of surgical procedures (Pavia & Andre, 2012; Liddle et al., 2015; Zhu et al., 2015), and health-related quality of life (HRQoL; Klassen et al., 2012; Antonarakis et al., 2013; Dimberg et al., 2015; Queiroz Herkrath et al., 2015). While these reviews are helpful, little consideration has been given as to how these different concepts and findings may interact and overlap. A more comprehensive picture of psychological adjustment to CL/P is necessary if we are to evaluate the current state of knowledge effectively and make advancements. Finally, it must be acknowledged that the field of CL/P has been making gradual shifts away from the traditional biomedical model and toward a more balanced, inclusive and patient-driven approach in recent years (Stock et al., *manuscript in preparation*). No existing review has effectively examined the literature within the context of these modern developments, or considered what this may mean for the field moving forward.

An authoritative, comprehensive, up-to-date and inclusive review of recent literature in the field of psychological adjustment to CL/P is currently lacking, and is clearly needed in light of the rapidly expanding and shifting field. The present article provides a narrative description of the literature published since the review carried out by Hunt et al. (2005), which was completed in December 2003.

Research Aims

- 1) To summarise the literature published between January 2004 and July 2015 according to five key domains of psychological adjustment, as well as overall reports of psychological wellbeing and quality of life (QoL)
- 2) To investigate the impact of contributing variables and potential associations between domains of adjustment
- 3) To assess the progress made toward advancing knowledge in this field during the last decade and to discuss future directions.

Method

Inclusion Criteria

All original, peer-reviewed articles pertaining to the psychological adjustment of individuals affected by CL/P published between January 2004 and July 2015 were included.

Quantitative, qualitative and mixed-methods papers were all considered. No age restrictions of the participants were imposed. Articles relating to all types of syndromic and non-syndromic CL/P were included. All methods of measurement, including self-reports, parent-reports and third-party reports (such as those obtained via clinicians, laypersons and teachers) were included. Articles published online while 'in press' were also included where available. Articles published in all languages were included where English translations could be obtained.

Exclusion Criteria

Case studies and unpublished dissertations were excluded. Articles relating to 'visible difference', 'disfigurement', 'craniofacial conditions' or similar search terms were excluded where results were not separated according to condition. 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. Articles relating only to the psychological adjustment of parents or other family members with CL/P were excluded, since this subject has been covered extensively in a previous review by Nelson and colleagues (2012). Articles regarding 'late presentation' for cleft repair in children, young people and adults were excluded since the findings were not comparable to routine treatment.

Search Strategy

The PsycInfo, MEDLINE, Science Direct, Web of Science and CINAHL databases were examined using similar but extended search terms to those detailed in the review conducted by Hunt et al. in 2005 (see Table 1 for a full list) [Table 1 near here]. The reference lists of included articles and previous reviews were checked to reduce the likelihood of any abstracts being missed. The literature search was performed by the first author between November 2014 and January 2015, and was updated again at the end of July 2015. Details of included and excluded articles are provided in Figure 1 [Figure 1 near here]. To assess quality control, the second author was given 15 randomly selected abstracts to review. The

evaluations made by the first and second authors were subsequently compared, and agreement was found to be high. Minor discrepancies were discussed until full agreement was reached.

Domains of Adjustment

In the context of this literature review, 'adjustment' was conceptualised as the process of the relative adaptation of an individual to the demands of their environmental context (see Seaton, 2009). The findings of each article were categorised according to five key domains of adjustment: Developmental Trajectory; Behaviour; Emotional Wellbeing; Social Experiences; and Satisfaction with Appearance and Treatment. In the absence of conventionally agreed domains of adjustment, the development of each domain was guided by recent literature (e.g. Feragen et al., 2015; Stock, Hammond, et al., in press) and designed to build upon and refine the categories used in the review by Hunt et al. (2005). Decisions regarding the definition and content of each domain was discussed and reflected upon by the authors throughout the review process. Table 2 describes the categorisation of findings into these key domains [Table 2 near here].

Results

Included Articles

In total, 148 articles were included in this narrative review. Of these articles, 128 were quantitative in nature. Sixteen studies used a qualitative approach, while four studies used a combination of both quantitative and qualitative measures. An overview of each article is described in more detail in Table 3 [Table 3 near here].

Summary of Findings across Key Domains of Adjustment

Initially, the findings pertaining to each domain are described in relation to how individuals with CL/P fare when likened to various comparison groups. Contributing variables, such as age, gender and cleft type are also described. Where appropriate, a description of associations across the different domains is presented. Finally, findings relating to overall psychological wellbeing or quality of life are summarised.

Developmental Trajectory

General Health

Aspects of general physical health were thought to impact upon psychological functioning. Individuals with CL/P were deemed to be in poorer general health in relation to comparison groups in two studies (Damiano et al., 2006; Knight, Cassell, Meyer & Strauss, 2015). According to one study, children with cleft palate only (CPO) were most likely to be defined as having special health care needs (Damiano et al., 2006). However, Eide, Skjærven, Irgens, Bjerkedal and Øyen (2006) did not find disability or mortality to be raised in those born with a cleft.

Physical characteristics, such as height (Nopoulos, Langbehn, Canady, Magnotta & Richman, 2007) were thought to be affected by CL/P, particularly in the case of CPO (Persson, Becker & Svensson, 2007). Weight was also found to be lower among individuals with CL/P (Persson et al., 2007; Smith, Walker, Badawi, Waters & MacLean, 2014), which was believed to be influenced in part by the number of respiratory events (Smith et al., 2014). Those with CPO were shown to have weaker muscular strength when compared to the reference group (Persson et al., 2007). Studies of sleep patterns in children with CL/P suggested no differences in one case (Brand et al., 2009) and an association with cognitive functioning in another (Smith et al., 2014).

Associated Conditions

A study by Bashir, Hodgkinson, Montgomery & Splitt (2008) reported a high percentage of 22Q11 diagnoses, predominantly among those with submucous cleft palate (smCP), and called for routine screening to be implemented. Burnell and colleagues (2014) found 16% of their sample to have a confirmed or strongly suspected syndromic condition, developmental disorder or further major malformation in addition to the cleft. Another 12.3% had mild developmental delay, while 3.7% had received a diagnosis of autism. According to Chetpakdeechit, Mohlin, Persson and Hagberg (2010), 25% of their sample had a diagnosed syndrome, 15.2% had Pierre Robin Sequence and 18% had an intellectual disability, the frequencies of which were influenced by the length of the cleft palate. In two studies of 10-year-old children, between 30-40% had a syndrome and/or a condition in addition to the cleft thought to impact on psychological adjustment, learning, development and/or cognition (Feragen & Stock, 2014; Feragen, Stock & Rumsey, 2014). Chetpakdeechit and colleagues (2010) also reported a number of affected organ systems. One study reported that children with CL/P were significantly more likely to have a developmental delay than the control group (Knight et al., 2015). Developmental difficulties seemed to be more prevalent among

types of cleft involving the palate (Chetpakdeechit et al., 2010; Burnell et al., 2014; Feragen et al., 2014).

Neurological Aspects

Neurological abnormalities were found among individuals with CL/P in a number of studies. Compared to controls, significant abnormalities and decreased volume and surface areas of the cerebrum and cerebellum were found in patients born with a cleft (Nopoulos et al., 2005; Boes et al., 2007; Nopoulos et al., 2007). The structure of the superior temporal plane was found to be disproportionately large (Shriver, Canady, Richman, Andreasen & Nopoulos, 2006) and abnormalities in the function of the distributed neural circuitry were also detected among adult men with CL/P compared to controls (Goldsberry, O'Leary, Hichwa & Nopoulos, 2006). According to Conrad, Canady, Richman and Nopoulos (2008), children with CL/P scored higher on tests of neurological soft signs, indicating non-specific cerebral dysfunction, particularly in younger participants (age range 7-17 years). Tissue distribution of cortical grey and white matter within the cerebrum were found to be abnormal in males, but were proportional to controls among girls with CL/P (Nopoulos et al., 2007). Among boys, right-sided clefts were associated with more abnormalities in brain structure, particularly in regard to reduced white matter volume (van der Plas, Conrad, Canady, Richman & Nopoulos, 2010). In another study, children with CL/P exhibited abnormally large cerebral cortex grey matter volumes, with decreased volumes of subcortical grey matter and cerebral white matter structures (Adamson, Anderson, Nopoulos, Seal & de Costa, 2014). However, no gender effects were found in this study.

Neurological abnormalities were related to lower scores on measures of intelligence (IQ; Shriver et al., 2006; Nopoulos et al., 2010), language (Goldsberry et al., 2006; Shriver et al., 2006) and reading (Goldsberry et al., 2006), as well as a generally impaired developmental trajectory (Nopoulos et al., 2007).

Cognitive Development and Language

One study suggested that individuals with CL/P have significantly lower general IQ than comparison groups (Hentges et al., 2011), while six studies found those with CL/P to score in line with comparison groups and/or within the normal range (Eide et al., 2006; Nopoulos et al., 2010; Conrad, McCoy, deVolder, Richman, & Nopoulos, 2014; Petrackova et al., 2015; Feragen & Stock, in press), except in the case of CPO (Persson, Becker & Svensson, 2008).

Individuals with CL/P demonstrated difficulties on tests of vocabulary (Scherer, Williams & Proctor-Williams, 2008; Young, Purcell & Ballard, 2010), grammar (Young et al., 2010), verbal IQ (Hentges et al., 2011; Conrad, Richman, Nopoulos & Dailey, 2009; Conrad et al., 2014), phonological awareness (Scheuerle, Guilford & Habal, 2004; Lee, Young, Liow & Purcell, 2015), narrative retelling (Klinto, Salameh & Lohmander, 2015) and receptive and expressive language (Scheuerle et al., 2004; Laasonen et al., 2004; Smith et al., 2014).

Varying degrees of dyspraxia/apraxia of oral motor mechanisms were also identified in one study (Laasonen et al., 2004). In addition, difficulties were identified in relation to performance IQ (Hentges et al., 2011), comprehension (Hentges et al., 2011), spelling (Hentges et al., 2011; Lee et al., 2015), visual memory (Richman, Wilgenbusch & Hall, 2005) and mathematics (Hentges et al., 2011; Wehby, Collett, Barron, Romitti, Ansley & Speltz, 2014). However, these results were not always replicated (Conrad et al., 2009; Collett, Leroux & Speltz, 2010; Young et al., 2012; Smith et al., 2014; Lee et al., 2015; Feragen, Særvold, Aukner & Stock, in press).

Potential problems with reading were reported in some studies (Collett, Stott-Miller, Kapp-Simon, Cunningham & Speltz, 2010), while not in others (Hentges et al., 2011; Feragen, Særvold et al., in press). According to one study, case versus control group differences on tests of reading were more apparent in those reporting higher socioeconomic status and among 'older' participants (age range 7-26 years; Conrad et al., 2014). Reading was also significantly associated with language skills (Feragen, Særvold et al., in press), verbal fluency, auditory memory (Conrad et al., 2014) and visual memory (Richman et al., 2005; Conrad et al., 2014). In one study, hearing did not affect reading test scores (Conrad et al., 2014), while differences in reading were reduced slightly when hearing problems were accounted for in another case (Collett, Stott-Miller et al., 2010).

In two studies, language development (Collett, Leroux et al., 2010) and babbling complexity (Scherer et al., 2008) of infants with CL/P was found to be significantly poorer than controls by the age of 12 months. Language development was not found to be affected by cleft type, articulation or hearing difficulties (Young et al., 2010). Three studies found individuals with CPO to have lower levels of cognitive functioning than the reference group (Laasonen et al., 2004; Persson et al., 2008; Feragen & Stock, in press). One study reported that those with more 'severe' clefts had lower performance IQ (Demir, Karacetin, Baghaki & Aydin, 2011). In studies where syndromes and additional conditions had already been identified, cognitive function was significantly affected by the presence of these conditions (Feragen, Borge &

Rumsey, 2009; Collett, Stott-Miller et al., 2010; Smith et al., 2014; Feragen & Stock, in press; Feragen, Særvold et al., in press).

Cognition scores were higher at 18 months of age in children whose mothers were more sensitive toward their infant, showed high rates of positive involvement and who looked at their infant more often (Murray et al., 2008). Further, verbal IQ at age seven years was mediated by maternal sensitivity at two months of age (Hentges et al., 2011). Mother-infant interaction, as well as demographic factors and early measures of mental and psychomotor development, were predictors of language outcomes at ages five and seven years (Collett, Leroux & Speltz, 2010).

Educational Experiences

Two studies reported individuals with CL/P to have lower educational achievement than their peers without CL/P (Wehby, Collett et al., 2014; Knight et al., 2015) and elevated rates of academic difficulties (Snyder & Pope, 2010), particularly in the case of CPO (Persson, Becker & Svensson, 2012). In contrast, three studies found no significant differences in educational achievement between those with and without CL/P (Cheung, Loh & Ho, 2007; van der Plas et al., 2013; Collett et al., 2014). According to five studies, young people with CL/P were more likely to be engaged in special educational services (Damiano et al., 2006; Collett, Leroux et al., 2010; Hentges et al., 2011; Wehby, Collett et al., 2014) and to repeat a grade (Lorot-Marchand et al., 2015) than their peers who were born without a cleft. This finding was particularly pronounced for those with palatal involvement (Damiano et al., 2006; Collett, Leroux et al., 2010; Collett et al., 2014; Wehby, Collett et al., 2014) and hearing difficulties (Tierney et al., 2015). In one sample of children with CL/P, 7% were placed in 'gifted' classes (Scheuerle et al., 2004).

Hearing difficulties, ear infections and OME were perceived to have a negative impact on learning at school in two qualitative studies (Tierney et al., 2015; Stock, Feragen & Rumsey, in press). The negative impact of children with CL/P missing more school than their classmates was also highlighted in three other studies (Chimruang et al., 2011; Knight et al., 2015; Lorot-Marchand et al., 2015). School performance was influenced by perceptions of general health in one study (Bos and Prah, 2011). In one qualitative study, a possible lack of understanding and support from teachers was also highlighted (Stock, Feragen, et al., in press).

Young people with CL/P scored lower than comparison groups on self-perceptions of 'general school' (Aravena, Gonzalez, Oyarzun, Coronado, in press), math and verbal ability (Gussy & Kilpatrick, 2006). However, five studies found no differences in relation to self-reports of overall scholastic competence among children with CL/P and comparison groups (Gussy & Kilpatrick, 2006; Oosterkamp et al., 2007; Gkantidis, Papamanou, Karamolegkou & Dorotheou, in press; Stock, Feragen & Rumsey, 2015), specifically in regard to females (Feragen, Stock & Kvalem, 2015). Boys were found to have higher ratings of scholastic competence than the reference group in one study (Feragen et al., 2015). In one qualitative study, participants reported having to work harder than their peers to demonstrate their capability (Stock, Feragen, et al., 2015). Perceptions of school were lower among minority ethnic groups (Broder, Wilson-Genderson & Sisco, 2012) and among adolescents with an additional condition (Feragen et al., 2015).

Employment

Three studies identified potential problems with stigma within the workplace (Chan, McPherson & Whitehill, 2006; Lorot-Marchand et al., 2015; Stock et al., 2015). However, adults generally believed that having CL/P had little or no influence over applying for a job (Oosterkamp et al., 2007) or their achievements at work (Oosterkamp et al., 2007; Stock et al., 2015). According to one qualitative study, some participants felt that having a visible difference had helped them to stand out within the workplace in a beneficial way (Stock et al., 2015).

Behaviour

Conduct Problems

In relation to comparison groups, individuals with CL/P reported higher levels of behavioural problems according to one study (Ha et al., 2013). No norms were available in one other study, but 38-44% of parents scored their child above the clinical cut-off in relation to behavioural conduct (Millar et al., 2013). However, Berger and Dalton (2009) found adolescents with CL/P to report fewer conduct problems than their peers without CL/P. In three other studies, no significant differences were found between individuals with CL/P and controls on measures of conduct problems (Murray et al., 2008; Brand et al., 2009; Collett, Cloonan, Speltz, Anderka & Werler, 2012).

One study found more behavioural problems in boys with CL/P compared to girls with CL/P at age 10 (Collet et al., 2012). However, when compared to a reference group, girls had similar conduct scores to the reference group, when boys had fewer conduct problems than the reference group (Feragen & Stock, in press). No differences in behavioural scores were found according to cleft type (Millar et al., 2013), while children with an additional condition reported significantly more conduct problems than children with a cleft only at age 10 (Feragen & Stock, 2014; Feragen & Stock, in press).

Behavioural problems were more likely if there was a history of CL/P in the family, if the child had been teased, or if the child had a visible scar (Hunt et al., 2007). Behavioural problems, inattention/hyperactivity and somatic symptoms were also associated with lower socioeconomic status (Wehby et al., 2012).

Internalising and Externalising Behaviours

Children were perceived to have higher levels of internalising behavioural problems according to parent (Hunt, Burden, Hepper, Stevenson & Johnston, 2007) and teacher (Murray et al., 2010) reports, and higher levels of externalising behavioural problems according to parent reports (Hunt et al., 2007). Males were more likely to score in the clinical range for aggressive behaviour than females, while females were more likely to score in the clinical range for anxious-depressed behaviours (Ha et al., 2013). Aggressive/oppositional behaviour was in line with normative samples in another study (Wehby et al., 2012). Feragen and Stock (in press) reported 10-year-olds with a cleft and an additional condition to have higher levels of withdrawal than in cases of a cleft only.

Inattention/Hyperactivity

No overall significant differences in measures of hyperactivity between those with CL/P and comparison groups were found in two studies (Brand et al., 2009; Feragen & Stock, in press), while the group with CL/P showed elevated rates of attention problems compared to norms in two other studies (Snyder and Pope, 2010; Conrad et al., 2014). In one study, as many as 18% of the sample had received a diagnosis of Attention Deficit and/or Hyperactivity Disorder (AD/HD; Richman, Ryan, Wilgenbusch & Millard, 2004), suggesting that AD/HD may be over-diagnosed in children with CL/P (Richman et al., 2004; Tierney et al., 2015). In contrast, three studies found similar or fewer problems with attention and hyperactivity in young people with a cleft compared to the reference group (Berger & Dalton, 2009; Wehby et

al., 2012; Feragen et al., 2015), and similar prevalence rates as in the general population (Feragen et al., 2014).

Two studies reported significantly higher scores of hyperactivity, impulsivity and inattention in males with CL/P than controls (Nopoulos et al., 2010; Ha et al., 2013), while another study found fewer hyperactivity/attention problems in boys compared to a reference group at age 10 (Feragen & Stock, in press) and 16 (Feragen et al., 2015). In one study, hyperactivity was found to be higher in children with CLP compared to those with CPO (Feragen & Stock, in press). Children with an additional condition reported significantly more attention problems than children with a cleft only at age 10 and at 16 years of age (Feragen & Stock, 2014; Feragen & Stock, in press; Feragen et al., 2015), a finding that was strongest in girls. However, levels of behavioural problems were still within the normal range.

Emotional Wellbeing

Psychiatric Conditions

Gourion and colleagues (2004) found a high incidence of CPO in patients with schizophrenia (30%) and their parents (24%). Major Depressive Disorder was also found to be significantly more prevalent in individuals with CL/P than in controls (Demir et al., 2011). The prevalence of psychiatric morbidity was 28.5% among patients with CL/P in one study, which was found to be significantly higher than the control group (Yunusa & Obembe, 2013). However, the rate of psychiatric diagnoses was not found to differ significantly from the general population in another study (Shriver et al., 2006). Psychopathology was related to lower performance IQ according to one study (Demir et al., 2011).

Emotional Functioning

A number of studies found individuals with CL/P to report poorer emotional functioning and more depressive symptoms than norms (Sinko et al., 2005; Hunt, Burden, Hepper, Stevenson & Johnston, 2006; Hunt et al., 2007; Mani, Carlsson & Marcusson, 2010), and more anxiety (Hunt et al., 2007) and somatic symptoms (Wehby et al., 2012). Twenty-four percent of 4-9-year-old children met the screening criteria for Separation Anxiety Disorder (SAD) according to parent reports in one study (Tyler, Wehby, Robbins & Damiano, 2013). SAD was found to be more common in non-Caucasian participants, and was related to lower parental education, poorer parental and child health status, children's problems with feeding and speech and lower satisfaction with facial appearance (Tyler et al., 2013). Other studies found individuals

with CL/P to report levels of emotional problems which were in line with (Gussy & Kilpatrick, 2006; Hunt et al., 2006; Oosterkamp et al., 2007; Brand et al., 2009; Wehby et al., 2012; Feragen & Stock, 2014; Gassling, Kessler et al., 2014; Lima et al., 2015), or lower than (Cheung, Loh & Ho, 2006; Munz, Edwards & Inglehart, 2011; Millar et al., 2013; Feragen et al., 2015) comparison groups. Two studies reported few children to score within the clinical range for depression at age 10 (Millar et al., 2013; Feragen & Stock, in press). Within one qualitative account, only a minority of participants attributed ongoing emotional difficulties to their CL/P (Stock et al., 2015).

In some cases, males with CL/P reported less emotional problems than females with CL/P (Eslami, Majidi, Aliakbarian & Hasanzadeh, 2013; Feragen & Stock, in press; Feragen et al., 2015), while the opposite was true according to two studies (Mani et al., 2010; Millar et al., 2013). Girls were more likely to report psychosomatic complaints within the clinical range (Ha et al., 2013). At age 10, girls with a visible cleft had fewer emotional difficulties than girls with a non-visible cleft according to parent reports, while the opposite was true for the boys (Feragen & Stock, in press). In contrast, males with a visible cleft reported significantly less depressive symptoms at age 16 than the comparison group in one study (Feragen, Kvaalem, Rumsey & Borge, 2010). At age 16, girls reported more emotional difficulties than the reference group, while boys reported fewer emotional difficulties compared to the reference group (Feragen et al., 2015). According to one study on adults, patients aged between 20 and 32 years of age were most negatively affected by emotional problems (Mani et al., 2010). A second study indicated that patients aged between 14-19 years had poorer emotional wellbeing than those aged 7-13 years (Broder et al., 2012). According to Bos and Prah (2011), children aged between eight and 12 had more emotional symptoms than those aged 13-15 years. In contrast, no differences were found in rates of depression or anxiety according to cleft type (Eslami et al., 2013; Millar et al., 2013), age (Eslami et al., 2013; Lima et al., 2015), gender or education (Lima et al., 2015). Lower emotional wellbeing scores were found among minority ethnic groups when compared to their White or Asian peers (Broder et al., 2012), and among children with an additional condition at age 10 (Feragen & Stock, 2014; Feragen & Stock, in press) and at age 16 according to parent reports (Feragen et al., 2015).

Several studies reported associations between emotional distress and other factors, including perceptions of general health (Bos & Prah, 2011), having a surgical recommendation, lower perceptions of school experiences (Broder et al., 2012; Broder, Wilson-Genderson, Sischo &

Norman, 2014), lower language and reading skills (Feragen, Særvold et al., in press) and overall HRQoL (Broder, Wilson-Genderson & Sischo, 2014).

Self-Concept

Self-concept scores were in line with norms and demonstrated no significant differences between individuals with CL/P and the control group according to one study (Boes et al., 2007). In another three studies, children with CL/P scored higher in relation to perceptions of self-image and/or physical ability than controls (Gussy & Kilpatrick, 2006; Pisula, Lukowska & Fudalej, 2014; Aravena et al., in press). However, one study found younger participants to have lower self-concept scores (Mani et al., 2010), while a qualitative study described young people as struggling with their self-image (Tiemens, Nicholas & Forrest, 2013).

Males scored lower than controls on ratings of personal power, while females scored higher than controls on measures of self-control (Pisula et al., 2014). A negative correlation between self-concept and mastery, and a positive association between depressive symptoms and mastery, were found in one study (Broder, Wilson-Genderson & Sischo, 2014). Another study asked children to draw themselves; all participants drew themselves with 'normal' mouths (Abd-Elseyed, Delgado & Livingstone, 2013).

Self-Esteem

In a study by Noor and Musa (2007), the majority of patients and their parents stated that self-confidence had been affected or "very much affected" by the cleft. Lower levels of self-esteem were also found among adults with CL/P (Cheung et al., 2007) and children with CL/P (Kramer, Gruber, Fialka, Sinikovic, & Schliephake, 2008) in relation to comparison groups. In contrast, self-esteem was found to be significantly higher in children with CL/P compared to their peers in two studies (Sagheri, Ravens-Sieberer, Braumann & von Mackensen, 2009; Millar et al., 2013). According to a number of studies, self-confidence could still be affected in adulthood (Havstam, Laakso, Lohmander & Ringsberg, 2011a; Hamlet & Harcourt, 2015; Stock et al., 2015). No effect of cleft type on self-esteem was found in one study (Millar et al., 2013).

Several studies reported associations between self-esteem and other psychological factors, such as coping styles (Pisula et al., 2014), recognition from peers (Chetpakdechit, Hallberg, Hagberg & Mohlin, 2009), less social anxiety and emotional distress (Cheung et al., 2007),

greater level of behavioural problems as perceived by the parents (Millar et al., 2013), and successful employment (Stock et al., 2015).

Social Experiences

Parent-Child Relationships

Mothers were found to be less sensitive toward their infant at the age of two months (Murray et al., 2008) and more disengaged (Despars et al., 2011; Montirosso et al., 2012) when compared to controls. Correspondingly, children with CL/P were found to be less engaged and to give fewer communicative signals (Frederickson, Chapman & Hardin-Jones, 2006; Montirosso et al., 2012). However, no differences were found between those born with CL/P and control groups in relation to the quality of attachment by 18 months of age in another (Murray et al., 2008). Children with insecure attachment were more likely to have problems with anxiety and depression than those who were secure, which was also evident in the control group (Murray et al., 2010). Maternal adjustment (Berger & Dalton, 2011; Despars et al., 2011) and a generally poorer parenting environment (Murray et al., 2010; Tyler et al., 2013; Stock, Feragen et al., in press) were believed to exacerbate difficulties with social relationships in later life.

Two studies suggested an effect of cleft type on the development of parent-child relationships. For children with a cleft lip, a significant association between maternal signs of depression and infant temperament was identified (Montirosso et al., 2012), while mothers of children born with more 'severe' types of cleft were less positively involved with their infant and looked at them less (Murray, 2008). However, the 'severity' of the cleft had no impact on attachment representations in another study (Despars et al., 2011). Having an antenatal diagnosis of CL/P was not related to mother-infant interaction at two months of age (Murray et al., 2008).

In contrast to these findings, other studies found mothers of children with CL/P to be more encouraging and to support their children more often than mothers of healthy children and those with other health conditions (Gassling, Christoph et al., 2014). Similarly, children with CL/P were found to demonstrate more autonomous behaviour and to interrupt their parent less frequently (Gassling, Christoph et al., 2014). According to self-reports, individuals with CL/P described normal or particularly close relationships with family members (Cheung et al., 2006; Gussy & Kilpatrick, 2006; Oosterkamp et al., 2007; Chimruang et al., 2011; Crerand et al., 2015; Gkantidis et al., in press), which was believed to act as a buffer against

social challenges (Hall, Gibson, James & Rodd, 2013; Tiemens et al., 2013; Stock, Feragen et al., in press). Nonetheless, two studies reported CL/P to have a negative impact on family life (Kramer et al., 2009; Gkantidis et al., in press). Ethnicity was also associated with greater family cohesion in one study (Crerand et al., 2015).

Social Functioning

Children with CL/P have been rated as being less socially motivated (van der Plas et al., 2013), less socially competent and as having poorer overall social functioning than their peers without CL/P (Slifer et al., 2004; Sinko et al., 2005; Frederickson et al., 2006; Boes et al., 2007; Kramer et al., 2009; Ha et al., 2013). In contrast, several studies found little or no influence of CL/P on social competence and functioning (Oosterkamp et al., 2007; Berger & Dalton, 2009; Munz et al., 2011; Collett et al., 2012; Gassling, Kessler, et al., 2014; Stock, Feragen, et al., 2015; Gkantidis et al., in press). In four studies, individuals with CL/P reported a fear of negative evaluation by others, anxiety or self-consciousness in social situations, and social withdrawal (Berger & Dalton, 2009; Murray et al., 2010; Chimruang et al., 2011; Demir et al., 2011; Stock et al., 2015). Higher levels of social avoidance and anxiety were found in adults with CL/P when compared to adolescents with CL/P and a control group in one study (Cheung et al., 2007). However, this finding was not replicated by van der Plas and colleagues (2013). Social Anxiety Disorder was found to be significantly more prevalent in individuals with CL/P than in controls in one study (Demir et al., 2011). In parallel, social anxiety was associated with poorer academic performance (van der Plas et al., 2013) and poorer social experiences based on self-reports (Berger & Dalton, 2009; Berger & Dalton, 2011). In contrast, a possible protective effect of close friendships was found in two studies (Berger & Dalton, 2009; Feragen et al., 2010), while the benefits of meeting another person with CL/P was discussed in three qualitative studies (Hamlet & Harcourt; Stock et al., 2015; Stock, Feragen et al., in press). Being socially competent was seen by patients as an important factor in overcoming social barriers in one qualitative study (Stock, Feragen et al., in press), while social withdrawal was related to poorer social experiences based on self-reports (Berger & Dalton, 2009; Berger & Dalton, 2011).

In two studies, social functioning was found to be associated with neurological abnormalities (Nopoulos et al., 2005; Boes et al., 2007). Two studies found the impact of CL/P on social functioning to decrease with age (Mani et al., 2010, age range 20-47 years; Gkantidis et al., in press, age range 9-33 years). Males with CL/P have been reported to have significantly

poorer peer relationships and social competency compared to norms (Boes et al., 2007; van der Plas et al., 2013; Feragen et al., 2015) and to be more likely to blame others as a coping strategy (Berger & Dalton, 2009). Females with CL/P were more likely to score within the clinical range for social problems (Ha et al., 2013), and females with a visible cleft provided slightly higher reports of peer problems than females with a non-visible cleft at age 16 (Feragen et al., 2015). However, reports of peer problems were still within the normal range for females with CL/P at both age 10 and 16 (Feragen & Stock; Feragen et al., 2015), while 16-year-old males with a visible cleft reported more positive perceptions of close friendships when compared to a reference group and when compared to those with a non-visible cleft (Feragen et al., 2015). Children with additional conditions reported more peer problems at age 10 compared to a reference group and when compared to children with a cleft only (Feragen & Stock, 2014; Feragen & Stock, in press). There was also a higher frequency of adolescents with additional conditions in the high risk group for social difficulties at age 16 (Feragen & Stock, in press).

Communication Difficulties

Articulation difficulties (Ruiter, Korsten-Meijer & Goorhuis-Brouwer, 2009) and hypernasality (Watterson, Macini, Brancamp & Lewis, 2013) were still present among school-aged children with CL/P, particularly in those with BCLP (Ruiter et al., 2009). Difficulties with being misunderstood created frustration and embarrassment in social situations (Damiano et al., 2006; Berger & Dalton, 2009; Berger & Dalton, 2011), and a more negative communication attitude (Havstam, Sandberg & Lohmander, 2011). Young people judged to be hypernasal were deemed to be less likely to make friends easily, less likely to fit in with friends and more likely to be teased (Watterson et al., 2013). Concurrently, speech problems, satisfaction with speech and/or reading difficulties were associated with social difficulties and general adjustment in a number of cases (Frederickson et al., 2006; Damiano et al., 2007; Murray et al., 2010; Berger & Dalton, 2011; Demir et al., 2011; Feragen, Særvold et al., in press; Gkantidis et al., in press). Forming an assessment of one's own speech, learning about one's communication and taking responsibility for communication were identified as important factors in coping with speech difficulties (Havstam, Laakson & Ringsberg, 2011). Hearing difficulties were also found to impair social functioning and to leave children feeling isolated (Tierney et al., 2015; Stock, Feragen et al., in press).

Social Reactions to CL/P

One study conducted in China found a general cultural bias in terms of a less favourable attitude towards disabled persons (Chan et al., 2006). When presented with a series of photographs of patients with CL/P, laypersons reported a high level of social distance (Pausch et al., in press). However, compared to results obtained 30 years earlier in a similar study, societal perceptions of patients with CL/P were believed to have improved (Pausch et al., in press). In a qualitative study, Hamlet and Harcourt (2015) found that older adults believed societal attitudes to appearance had changed, but not necessarily for the better. In one qualitative study, public perceptions of CL/P were found to have an impact on psychological adjustment (Stock, Feragen et al., in press). Three eye-tracking studies found that laypersons are more often fixated on the mouths of patients with CL/P and spend longer looking at the areas of the face affected by CL/P (Meyer-Marcotty, Gerdes, Reuther, Stellzig-Eisenhauer & Alpers, 2010; Meyer-Marcotty, Gerdes, Stellzig-Eisenhauer & Alpers, 2011; van Schijndel, Litschel, Maal, Berge & Tasman, in press). However, this was not related to laypersons' ratings of negative personality traits (van Schijndel et al., in press).

Reports of Teasing

Included studies showed wide variations in the reported frequency of teasing among patients with CL/P as rated by patients and parents, ranging from 20% to 75% (Semb et al., 2005; Hunt et al., 2006; Hunt et al., 2007; Noor & Musa, 2007; Feragen et al., 2009; Feragen & Borge, 2010; van Lierde et al., 2012; Lorot-Marchand et al., 2015; Feragen, Særvold et al., in press). Nineteen out of 20 participants reported that they had been teased about appearance or speech in another study (Demir et al., 2011). In a minority of cases, patients reported having been physically hurt (Hunt et al., 2006). Perceived teasing was reported to be higher among patients with CL/P than among controls (Hunt et al., 2006; Hunt et al., 2007; Demir et al., 2011; van Lierde et al., 2012). In one qualitative study, the majority of patients reported being teased at some point, although their perceptions of the severity and consequences of teasing varied considerably (Stock, Feragen et al., in press). This finding was supported by one quantitative study, which found that not everyone who reported teasing confirmed that the teasing worried them (Noor & Musa, 2007).

Teasing was predominantly related to appearance (lip, nose, teeth) or speech (Semb et al., 2005; Hunt et al., 2006; Hunt et al., 2007; Noor & Musa, 2007; Berger & Dalton, 2009; Demir et al., 2011; van Lierde et al., 2012; Lorot-Marchand et al., 2015). In one qualitative study (Tiemens et al., 2013), participants reported that their cleft lip had made them feel more

shy and less confident. The use of social withdrawal and distraction as coping strategies were related to poorer social experiences and subsequently to poorer adjustment (Berger & Dalton, 2011). Teasing was reported to occur at school in most cases (van Lierde et al., 2012; Lorot-Marchand et al., 2015), although two studies also reported perceived discrimination to also occur in the workplace (Lorot-Marchand et al., 2015; Stock et al., 2015). In a study by Semb and colleagues (2005), the majority of teasing was found to occur between the ages of 8-11 years, followed by 12-15-year-olds, with very little teasing occurring before or after this. However, Noor and Musa (2007) reported teasing to occur as early as two years of age, which persisted for a minority of individuals with CL/P until the age of 17 years. The highest onset of teasing in this study was seven years of age (Noor & Musa, 2007). Two studies reported high levels of teasing during adolescence (Berger & Dalton, 2009; Lorot-Marchand et al., 2015).

Several studies reported associations between teasing and other psychological factors. A history of teasing/bullying was a significant predictor of psychological difficulties and internalising/externalising behaviour problems, more so than having CL/P per se (Hunt et al., 2006). Further, peer harassment was related to appearance dissatisfaction at ages 10 and 16 (Feragen & Borge, 2010). Another study showed an association between the visibility of the cleft and appearance satisfaction that was fully mediated by reported peer harassment (Feragen & Borge, 2010). However, one study found no differences between those who reported taunting and those who did not in relation to satisfaction with appearance (Lorot-Marchand et al., 2015). Teasing over appearance, but not speech, was significantly related to depressive symptoms in two studies (Demir et al., 2011; Lorot-Marchand et al., 2015), while teasing was correlated with poorer language and reading skills in another (Feragen, Særvold et al., in press). In contrast, absence of perceived teasing was associated with evidence of psychosocial resilience in one study (Feragen et al., 2009).

Social Acceptance

Individuals born with CL/P reported not being aware of their difference until late childhood (Chetpakdechit et al., 2009). How they were treated by others played a key role subsequently in shaping their own perceptions of their cleft (Havstam, Laakso & Ringsberg, 2011). Perceived stigma or unwanted interest from others, including staring, comments and questions, was reported by parents and patients in a number of studies (Berger & Dalton, 2009; Chetpakdechit et al., 2009; Feragen et al., 2009; Alansari, Bedos & Allison, 2014;

Tiemens et al., 2013; Stock & Rumsey, 2015). Individuals with CL/P believed these experiences to have made them more vulnerable to feeling different from their peers (Chetpakdeechit et al., 2009; Havstam, Laakso & Ringsberg, 2011; Alansari et al., 2014; Tiemens et al., 2013). In two qualitative studies, participants struggled with wanting to feel good about their difference and to not care about what others thought, despite wanting to feel normal and to be accepted (Havstam, Laakso & Ringsberg, 2011; Tiemens et al., 2013). Teasing (Havstam, Laakso & Ringsberg, 2011) and problems with nasality (Watterson et al., 2013) posed a threat to feelings of normality and ratings of social acceptance. Some individuals with CL/P reported that they lacked recognition (Chetpakdeechit et al., 2009) and felt devalued by others (Tiemens et al., 2013). In turn, low levels of perceived social acceptance were found to be associated with gaze avoidance and less assertive social patterns (Slifer et al., 2004; Slifer et al., 2006). Feelings of social isolation were reported in one study (Hamlet & Harcourt, 2015). However, the study by Slifer and colleagues (2006) did not find any differences in social acceptance scores between individuals with CL/P and their peers without CL/P. In a qualitative study, perceptions of difference varied from “not being different” to being different in a negative or positive light (Stock, Feragen et al., in press). Similarly, some participants felt that their cleft was not very noticeable to other people, while others felt that having CL/P made them stand out, either in a negative or positive way (Stock, Feragen et al., in press).

Social acceptance scores were also be affected by gender, age and cleft visibility. Males with CL/P reported more positive perceptions of social acceptance at age 10 when compared to a reference group (Feragen et al., 2010; Feragen & Stock, in press), while males with a visible cleft reported more positive perceptions of social acceptance than both the reference group and those with a non-visible cleft at age 16 (Feragen et al., 2010; Feragen et al., 2015). In a study with older adults, Hamlet and Harcourt (2015) identified a tension between not wanting to be noticed by others because of their CL/P and feeling ignored because of their older age. Other factors with the potential to impact upon social acceptance included sexual orientation and cultural factors (Stock, Feragen et al., in press).

Romantic Relationships

Compared to norms, individuals with CL/P had higher (Gussy & Kilpatrick, 2006) or equal (Feragen et al., 2015) self-concept scores in regard to romantic relationships. In a qualitative study, many adults did not believe that CL/P had had a negative effect on their intimate

relationships overall, while some felt it had had a positive influence (Stock et al., 2015). In this case, struggles with romantic relationships were predominantly related to young adulthood (Stock et al., 2015). In one study (Havstam, Laakso & Ringsberg, 2011), participants had waited longer than most of their friends to have their first romantic partner, while worries about romantic relationships were based on the belief that relationships were conceptualised as confirmation of attractiveness. Males with CL/P reported more positive perceptions of romantic appeal than females with CL/P in one study, although both males and females with CL/P still reported higher perceptions of romantic appeal than norms (Feragen et al., 2015).

Satisfaction with Appearance and Treatment

Patient and Parent Satisfaction with Appearance

Eighteen studies investigated general satisfaction with appearance as reported by patients and/or their parents. In spite of a reasonable level of satisfaction being reported among patients and their parents (van Lierde et al., 2012; Wehby et al., 2012; Tyler et al., 2013), lower levels of satisfaction with facial appearance were found when compared to a control or reference group (Slifer et al., 2004; Hunt et al., 2006; Hunt et al., 2007; Chuo et al., 2008; Versnel, Duivenvoorden, Passchier & Mathijssen, 2010; van Lierde et al., 2012; Wehby et al., 2012). The appearance of the nose, upper lip, facial profile and teeth were often cited as areas of dissatisfaction (Semb et al., 2005; Sinko et al., 2005; Hunt et al., 2006; Hunt et al., 2007; Oosterkamp et al., 2007; Chuo et al., 2008; Berger & Dalton, 2009; Meyer-Marcotty & Stellzig-Eisenhauer, 2009; van Lierde et al., 2012; Nkenke, Stelzle, Vairaktaris & Knipfer, 2013; Byrne, Chan & O'Broin, 2014). Damiano et al. (2006) found satisfaction with appearance to be higher in patients with CL/P than in patients with clubfoot, while 57% of patients in a study by Lorot-Marchand et al. (2015) reported that they found their face beautiful. When asked about their appearance, 13% of patients in a study by Meyer-Marcotty and Stellzig-Eisenhauer (2009) indicated that 'everything was OK'. Patients reported significantly higher satisfaction with facial features not associated with CL/P (e.g. ears, eyes, hair) and with overall appearance than reference groups (Gussy & Kilpatrick, 2006; Berger & Dalton, 2009). This finding was also replicated in a qualitative study by Gkantidis et al. (in press).

In several studies, satisfaction with appearance was examined according to age and gender. In several cases, males were found to be more dissatisfied with appearance than females

(Semb et al., 2005; Landsberger et al., 2006; Hunt et al., 2007; Byrne et al., 2014). Contrastingly, the opposite was indicated in two other studies (Sinko et al., 2005; Mani, Reiser, Andlin-Sobocki, Skoog & Holmström, 2013). Four studies found no effect of gender (Hunt et al., 2006; Oosterkamp et al., 2007; Pitak-Arnop, Hemprich, Dhanuthai, Yildirim & Pausch, 2011; Feragen et al., 2015). In two cross-sectional studies, satisfaction with appearance was found to decrease with age (Hunt et al., 2006, age range 8-21 years; Gkantidis et al., in press, age range 9-33 years), while one study identified this effect in female patients only (Byrne et al., 2014, age range 14-53 years). Patient ratings of appearance were found to be lowest in females aged between 24 and 30 years in one study (Sinko et al., 2005). The age of the patient did not affect satisfaction with appearance in two studies (Oosterkamp et al., 2007; Pitak-Arnop et al., 2011). Two qualitative studies indicated that satisfaction with appearance may fluctuate over time, and continues to be an important factor during adulthood and older age (Hamlet & Harcourt, 2015; Stock, Feragen et al., in press).

Satisfaction with appearance was found to be significantly lower in those patients with a BCLP when compared to those patients born with other types of CL/P (Oosterkamp et al., 2007). Having a visible cleft was associated with lower overall satisfaction with appearance in three studies (Hunt et al., 2006; Feragen et al., 2015; Gkantidis et al., in press), and with lower satisfaction with cleft-related facial features in two others (Feragen & Stock, in press; Feragen et al., 2015). Patients with CPO were found to be more satisfied with the appearance of the nose and lip than those with a visible cleft (Gkantidis et al., in press). Having a condition in addition to the cleft was associated with lower overall satisfaction with appearance at age 10 (Feragen & Stock, in press), but not at age 16 (Feragen et al., 2015), although the effect size was small. Satisfaction with appearance was also found to be significantly lower in South Asian participants when compared to Caucasian participants, particularly in relation to the appearance of the nose (Reekie, 2011). A larger number of surgeries was related to greater satisfaction with appearance in two studies (Demir et al., 2011; Mani et al., 2013), but not in one other (Semb et al., 2005).

Ratings of physical appearance were significantly associated with QoL (Damiano et al., 2007; Mani et al., 2013), general adjustment (Berger & Dalton, 2011), professional life (Gkantidis, Papamanou, Christou & Topouzelis, 2013), social experiences (Feragen et al., 2009; Feragen & Borge, 2010; Murray et al., 2010; Versnel et al., 2010; Gkantidis et al., 2013), mental health and vitality (Oosterkamp et al., 2007), self-esteem (Versnel et al., 2010; Millar et al., 2013), behavioural problems (Wehby et al., 2012; Millar et al., 2013) and withdrawn, anxious

and/or depressive symptoms (Feragen et al., 2010; Murray et al., 2010; Millar et al., 2013). Further, subjective ratings of appearance mediated the relationship between social experiences and depressive symptoms (Feragen et al., 2010).

Perceptions of Appearance among Professionals and Laypersons

Sixteen studies asked laypersons, as well as specialist and non-specialist professionals, to assess patient photographs on a range of different measures of aesthetics. Within these studies, patients with CL/P were frequently rated more negatively than controls in relation to facial appearance and symmetry (Meyer-Marcotty, Alpers, Gerdes & Stellzig-Eisenhauer, 2010; Pitak-Arnop et al., 2011; Meyer-Marcotty, Kochel, et al., 2011; Eichenberger, Staudt, Pandis, Gnoinski & Eliades, 2014). Professionals and laypersons highlighted the mid-face region, the nose, the teeth and the upper lip as causing aesthetic impairment (Chetpakdeechit et al., 2011; Ferrari Júnior, Ayub, Capelozza Filho, Pereira Lauris & Garib, 2015). In two studies, patients with CL/P were also perceived to present more negative facial expressions than controls (Meyer-Marcotty, Gerdes, Reuther, et al., 2010; Meyer-Marcotty, Alpers, et al., 2010). Greater objective asymmetry of the face was associated with lower subjective ratings of appearance in one study (Meyer-Marcotty, Gerdes, Reuther, et al., 2010). In one study, males with CL/P were rated as being less attractive than females with a cleft according to laypersons (Meyer-Marcotty, Gerdes, et al., 2011).

Fourteen studies compared perceptions of treatment outcomes among patients, laypersons, and specialist and non-specialist professionals. One study found patients' ratings of their own appearance to be lower than ratings provided by professionals (Sinko et al., 2005). In contrast, patients gave more positive ratings of their own appearance when compared to professionals' ratings in three cases (Hens et al., 2011; Gkantidis et al., 2013; Byrne et al., 2014). In an analysis of specific facial features, Meyer-Marcotty and Stellzig-Eisenhauer (2009) found patients to be less satisfied with the nose and lip, while professionals were more concerned with the teeth.

There was a reasonable level of agreement between professionals and laypersons in relation to ratings of appearance in most cases (Meyer-Marcotty & Stellzig-Eisenhauer, 2009; Pitak-Arnop et al., 2011; Papamanou, Gkantidis, Topouzelis & Christou, 2012; Gkantidis et al., 2013; Watterson et al., 2013), with some exceptions. In one study, professionals rated patients as being significantly less attractive in all components of the face than did laypersons (Foo, Sampson, Roberts, Jamieson & David, 2013), whereas in three studies, professionals

gave higher ratings of appearance than those provided by laypersons (Chetpakdeecheit et al., 2011; Eichenberger et al., 2014; Ferrari Júnior et al., 2015). In four studies, laypersons were found to be the least satisfied group in relation to patients' appearance (Papamanou et al., 2012; Gkantidis et al., 2013; Foo et al., 2013; Eichenberger et al., 2014). Despite not always being satisfied with the treatment outcome, raters tended to report that patients' aesthetics were 'acceptable' overall (Ferrari Júnior et al., 2015; Gkantidis et al., 2015).

Objective Assessment of Aesthetics

Four studies used 3-Dimensional imaging to assess facial aesthetics objectively. These studies demonstrated patients with CL/P to have significantly more asymmetry of the face than controls (Meyer-Marcotty, Gerdes et al., 2010; Meyer-Marcotty, Gerdes et al., 2011), with the mid-face region showing the most asymmetry (Meyer-Marcotty & Stellzig-Eisenhauer, 2009; Meyer-Marcotty, Gerdes et al., 2010). However, few clear links were found between objective assessments of appearance and subject evaluations (Semb et al., 2005; Meyer-Marcotty & Stellzig-Eisenhauer, 2009; Millar et al., 2013).

Satisfaction with Function

Satisfaction with function was examined in 12 studies. A variety of functional impairments were reported, including low speech intelligibility, problems with resonance, difficulties with nasal breathing, poor oral health, hearing difficulties, Otitis Media with Effusion (OME), problems with eating and drinking and reduced overall wellbeing (Oosterkamp et al., 2007; Chuo et al., 2008; Ruiter et al., 2009; Collett, Stott-Miller et al., 2010; Bos & Prahl, 2011; Chimruang et al., 2011; van Lierde et al., 2012; Wehby et al., 2012; Tyler et al., 2013; Knight et al., 2015; Tierney et al., 2015; Aravena et al., in press; Feragen, Særvold et al., in press; Gkantidis et al., in press). In contrast, two studies found levels of satisfaction with function to be high overall (Munz et al., 2011; Gkantidis et al., in press). Few differences in the ratings given by patients, laypersons and professionals were found (Sinko et al., 2005; Brunnegård, Lohmander & van Doorn, 2009), although only 7% of the variance in subjective satisfaction with speech was explained by clinicians' assessments in one study (Feragen, Særvold et al., in press).

In a sample of 8-21-year-olds, being older was associated with dissatisfaction with speech (Hunt et al., 2006). In contrast, younger participants (age 8-12 years) reported more symptoms related to functional difficulties (Bos & Prahl, 2011). Satisfaction with function was found to be significantly lower in those patients with a BCLP when compared to those

patients born with other types of CL/P (Oosterkamp et al., 2007). Satisfaction with functional outcomes was also found to be significantly lower in South Asian participants when compared to Caucasian participants, especially in relation to speech (Reekie, 2011).

Motivations for Surgical Intervention

Patients reported wanting further treatment in several studies (Semb et al., 2005; Hunt et al., 2007; Lorot-Marchand et al., 2015), with nose and lip corrections being the most sought-after procedures (Semb et al., 2005; Sinko et al., 2005; Chuo et al., 2008; Lorot-Marchand et al., 2015). Motivations for undergoing further treatment included dissatisfaction with appearance (Augsornwan, Namedang, Pongpagatip & Surakunprapha, 2011; Tiemens et al., 2013; Byrne et al., 2014; Lorot-Marchand et al., 2015), low self-perceptions (Alansari et al., 2013; Tiemens et al., 2013), a desire to improve speech (Sharp et al., 2008), a desire to reduce nasal obstruction (Hens et al., 2011) and social benefits (Sharp et al., 2008; Augsornwan et al., 2011; Tiemens et al., 2013). However, patients, professionals and laypersons frequently disagreed on whether further treatment was necessary. In contrast to the findings above, patients did not often believe additional treatment was needed, despite professionals and laypersons believing patients would benefit (Sinko et al., 2005; Meyer-Marcotty & Stellzig-Eisenhauer, 2009; Foo et al., 2013; Nkenke et al., 2013; Stock et al., 2015).

A number of other studies described reasons for treatment refusal, which included patients' reports of feeling satisfied with treatment, having positive self-perceptions, being able to cope, believing no more could be done, a fear of the treatment failing or being tired of treatment (Havstam, Laakso & Ringsberg, 2011; Hall et al., 2012; Alansari et al., 2013; Stock et al., 2015). Patients were more likely to refuse treatment, or to find the decision-making process more difficult if the procedure was for cosmetic reasons only (Havstam, Laakso & Ringsberg, 2011; Hall, Gibson, James & Rodd, 2012). Some patients had also declined treatment due to a fear of the treatment failing (Hall et al., 2012; Stock et al., 2015).

Managing patients' expectations of treatment was considered to be an important factor in the decision-making process (Stock, Feragen et al., in press). Experiences of treatment during childhood shaped patients' decision process when considering optional surgeries offered at a later stage (Alansari et al., 2013; Stock, Feragen et al., in press). In a sample of 8-17-year-olds, young people were found to adopt their own measures to improve appearance, including wearing make-up and nice clothes (Hall et al., 2012). Some only took photographs from their

'best side' and some consciously smiled more to make their cleft less noticeable (Hall et al., 2012).

Experiences of Treatment

Eight studies described patients as feeling nervous, intimidated by and/or frightened of treatment and clinic appointments (Noor & Musa, 2007; Luoto, Lahti, Nevanpera, Tolvanen & Locker, 2008; Bos & Prahl, 2011; Vogels, Aartman & Veerkamp, 2011; Hall et al., 2012; Dogan, Serin, Uzel & Seydaoglu, 2013; Krikken et al., 2015; Tierney et al., 2015), while others reported that treatment could be burdensome (Chetpakdechit et al., 2009; Havstam, Laakso & Ringsberg, 2011; Alansari et al., 2013). Females reported more concern about treatment than males in two studies (Luoto et al., 2008; Bos & Prahl, 2011). Treatment anxiety was highest in children aged four to six years (Vogels et al., 2011), but was generally found to reduce with age and with previous experience of treatment (Luoto et al., 2008; Dogan et al., 2013; Krikken et al., 2015). Some patients felt it was difficult for them to discuss concerns with specialists, while others reported feeling more confident in expressing their views (Noor & Musa, 2007; Hall et al., 2012). Patients raised concerns about feeling pressured to comply with treatment recommendations and/or having a lack of influence over treatment decisions (Noor & Musa, 2007; Havstam, Laakso & Ringsberg, 2011; Hall et al., 2012; Stock et al., 2015). Concurrently, the importance of treatment autonomy and involvement was discussed in a number of studies (Semb et al., 2005; Noor & Musa, 2007; Chuo et al., 2008; Hall et al., 2012; Stock, Feragen et al., in press). Some patients reported that they found clinic information difficult to understand (Noor & Musa, 2007), and felt that information provision and treatment referrals could be improved (Augsornwan et al., 2011; Hamlet & Harcourt, 2015; Stock & Rumsey, 2015; Stock et al., 2015). Treatment for CL/P was often described as a process, rather than an event (Hall et al., 2013; Alansari et al., 2013; Gkantidis et al., 2013).

Treatment experiences were associated with a wide range of factors. Hospital appointments and undergoing treatment affected feelings of normality and social acceptance according to one study (Havstam, Laakso & Ringsberg, 2011). Patients who reported fear of treatment reported the use of more coping skills (Vogels et al., 2011) and poorer emotional and social wellbeing (Luoto et al., 2008). Children with more episodes of treatment reported a more positive mental state in two studies (Luoto et al., 2008; Demir et al., 2011), while a higher number of surgeries was associated with increased anxiety, depression, somatic symptoms

(Wehby et al., 2012), lower overall emotional wellbeing and poorer HRQoL (Broder et al., 2012) according to two other studies. In contrast, one study reported that satisfaction with treatment was not correlated with patients' social or psychological QoL (Munz et al., 2011). Low socioeconomic status was described as a barrier to attending treatment appointments in two studies (Smillie, Yong, Harris, Wynne & Russell, 2015; Stock, Feragen et al., in press).

Satisfaction following Medical Intervention

Thirteen studies investigated parent and patient satisfaction with treatment outcome following completion of treatment or after a surgical intervention. Overall, a high level of parent and patient satisfaction with treatment outcome was described (Semb et al., 2005; Damiano et al., 2006; Noor & Musa, 2007; Hens et al., 2011; Munz et al., 2011; Pitak-Arnop et al., 2011; Byrne et al., 2014; Stock et al., 2015). Patients reported that their appearance (Sandor & Ylikontiola, 2006; Sharp et al., 2008; Hens et al., 2011; Byrne et al., 2014) and function (Sharp et al., 2008; Hens et al., 2011) had improved as a result of treatment, that they would undergo the treatment again knowing the result (Sandor & Ylikontiola, 2006; Hens et al., 2011; Byrne et al., 2014), and that they would recommend the treatment or procedure to others (Sharp et al., 2008). Professionals were also satisfied with post-operative appearance on the whole (Byrne et al., 2014). However, a minority of patients and professionals stated they were 'uncertain', believed there to be 'no improvement', or felt that appearance and/or function had 'worsened' (Noor & Musa, 2007; Hens et al., 2011; Byrne et al., 2014). Two qualitative studies reported that treatment did not always yield results which outweighed the burden associated with treatment (Alansari et al., 2013; Stock et al., 2015). Satisfaction with treatment varied according to the type of procedure performed in three cases (Cheung et al., 2006; Landsberger et al., 2006; Pitak-Arnop et al., 2011), and with the amount of pain experienced in two studies (Munz et al., 2011; Alansari et al., 2013). Three studies found discrepancies in satisfaction with appearance and treatment outcomes between patient and parent reports. In some cases, the parents reported being more satisfied with the patient's treatment outcomes than the patients themselves (Berger & Dalton, 2009; Munz et al., 2011). Five other studies found moderate agreement between patients and parents (Semb et al., 2005; Noor & Musa, 2007; van Lierde et al., 2012; Gkantidis et al., 2013; Gkantidis et al., in press).

Timing of Surgery

According to parent reports, children who had their cleft lip repaired within a few days of birth scored more highly on measures of self-esteem than those children who had their cleft

lip repaired at three to six months of age (Petráčkova et al., 2015). Patients undergoing cleft repair at three to four months of age scored significantly lower on tests of verbal IQ, full IQ and grammar compared to those undergoing cleft repair neonatally (Hentges et al., 2011), while IQ was not related to timing of repair in another study (Petrackova et al., 2015). Additionally, undergoing cleft repair at three to four months of age was related to poorer cognitive development, while those undergoing neonatal repair had cognitive scores which were comparable to controls (Murray et al., 2008).

General Adjustment and Wellbeing

Quality of Life and Health-Related Quality of Life

According to five studies, the influence of CL/P on total QoL was found to be low (Kramer et al., 2008; Augsornwan et al., 2011; Smith et al., 2014; Stock et al., 2015; Aravena et al., in press). No differences were found in relation to QoL scores according to cleft type (Kramer et al., 2008). Overall QoL was related to self-concept, self-perceptions of mastery (Broder, Wilson-Genderson & Sischo, 2014), self-esteem and coping styles (Pisula et al., 2014). Differences between patient and parent reports were found in one study, with higher levels of QoL being reported by patients (Kramer et al., 2008). No differences were found between individuals with CL/P and controls in relation to total HRQoL scores in three studies (Oosterkamp et al., 2007; Sagheri et al., 2009; Mani et al., 2010). Younger children with CLP (2-7 years) had higher HRQoL scores than same-aged children with CPO, although this pattern was reversed for older children (8-12 years; Damiano et al., 2007).

General Adjustment

Individuals with CL/P did not score differently to norms in terms of overall psychological adjustment according to two studies (Berger & Dalton, 2009; Smith et al., 2014). According to one study, nineteen percent of adolescents reported that they had experienced no problems relating to their cleft (Berger & Dalton, 2009). Differences between patient and parent reports were found in one study, with higher adjustment scores being reported by patients (Berger & Dalton, 2009). Boys with CL/P reported greater adjustment difficulties than girls with a cleft, although scores were still within the normal range, irrespective of age and cleft type (Berger & Dalton, 2009). According to another study, children with a non-visible cleft had significantly fewer adjustment problems than those with a visible cleft according to one other, while children with an additional condition were found to have significantly more

psychological difficulties than those with a cleft only, although scores were within the normal range for all subgroups (Feragen & Stock, in press).

The decision to have children

Four studies investigated the influence of psychological adjustment to CL/P on adults' decision to have children. In one study, childlessness was higher among women born with CL/P compared to women in the general population, especially among women with a CPO (Yttri, Christensen, Knudsen & Bille, 2011). The average age for women with CL/P to have their first child was also older than women without CL/P. However, if women with CL/P did have children, there was no significant difference in regard to the number of children (Yttri et al., 2010). In one study, having an affected first child was a better predictor of future fertility rates than the mother being affected with CL/P herself (Wehby, Nyarko & Murray, 2014). Concerns regarding the recurrence risk of CL/P were mentioned by three studies (O'Hanlon, Camic & Shearer, 2012; Stock & Rumsey, 2015; Stock et al., 2015). However, O'Hanlon and colleagues (2012) found few significant differences in adjustment between parents with and without a diagnosis of CL/P, and Stock and Rumsey (2015) reported a number of beneficial outcomes, including the potential to create a positive cycle of adjustment among generations.

Discussion

Synthesis of Findings

In this narrative review of the literature, findings from studies pertaining to the psychological adjustment of individuals with CL/P and published during the last decade (Jan 2004-July 2015) were presented. Findings were categorised according to five key domains: 'Developmental Trajectory', 'Behaviour', 'Emotional Wellbeing', 'Social Experiences' and 'Satisfaction with Appearance and Treatment'. Contributing variables, such as gender, age, ethnicity, cleft type, the presence of additional conditions and socioeconomic status were also identified. In addition, findings relating to overall QoL, HRQoL and general psychological wellbeing were provided. Finally, associations between domains of adjustment were examined.

In 2005, the comprehensive review published by Hunt and colleagues concluded that while the majority of individuals born with CL/P do not appear to experience major psychological problems, some specific problems in relation to satisfaction with appearance, behaviour,

depression and anxiety may arise. In addition, reports of self-concept, satisfaction with appearance, depression, attachment, learning problems and interpersonal relationships were thought to vary by cleft type. These conclusions will now be discussed in relation to the findings of the current review.

In the present review, dissatisfaction with appearance seemed to be a prevalent concern. However, this concern may actually be comparable to, or better than the levels of appearance dissatisfaction being reported among the general population, and elevated only in relation to the facial features affected by the cleft. This is an interesting development for the field and calls into question the tendency for studies to assess satisfaction with appearance predominantly in relation to treatment. These findings suggest the need to distinguish between perceptions of general physical appearance and aspects of appearance integral to CL/P. Satisfaction with appearance is also known to vary according to age and gender in both the general population and among individuals with visible conditions (Smolak, 2004; Rumsey & Harcourt, 2012); factors which are difficult to decipher without large and age-appropriate samples, in addition to appropriate reference or control groups. What is clear is that dissatisfaction with appearance can impact upon many domains of psychological functioning, and therefore a clearer understanding of the factors which contribute to the development and maintenance of appearance concern in this population is needed. The ways in which cleft treatment to improve aesthetics interacts with these relationships also requires further consideration. Motivations for and expectations of treatment were highlighted in the present review, along with patient perceptions of treatment autonomy and relationships with health professionals.

Fewer papers evaluating the behaviour of those born with CL/P appear to have been published since the review by Hunt et al. (2005). When overall behavioural conduct is measured, few difficulties are found. However, high levels of internalising/externalising behaviours, as well as elevated levels of inattention/hyperactivity have been reported. It could be that behavioural difficulties are in fact an expression of underlying emotional, cognitive or neurological problems, and/or related to the presence of an additional condition. Four studies proposed that behavioural difficulties may be misdiagnosed among individuals with CL/P. In addition, the relatively small number of studies included in this domain in the current review could suggest a shift in the focus of recent research and/or illustrates that the way in which domains of adjustment are conceptualised affects the decision of where to place the research findings. This review therefore raises an important consideration in relation to how behavioural and

psychological problems are conceptualised, assessed and reported in both clinical practice and within research samples.

The current review does not lend full support to the previous finding that levels of anxiety and depression are raised among individuals with CL/P (Hunt et al., 2005). Rather, the picture appears to be more complex and affected by numerous confounders. This discussion is also relevant in regard to studies investigating self-concept and self-esteem. However, a number of recent studies have begun to investigate associations between emotional functioning and other domains of adjustment, such as positive and negative social experiences. This seems to be a positive step forward for the field in terms of understanding whether the cleft, or other factors, are responsible for emotional distress and poorer self-perceptions, and could help to identify the subgroups which may be most vulnerable.

The current review presents a more thorough investigation of cognitive functioning than the earlier review by Hunt et al., (2005), and may reflect an increase in interest in this area. The previously reported prevalence of learning problems among individuals with CL/P was confirmed, and although general IQ does not seem to be affected, some specific deficits in cognitive and language development have been repeatedly demonstrated. However, these findings are likely affected by the presence of additional conditions and/or neurological abnormalities demonstrated by recent investigations. In addition, some consensus that outcomes may be most affected in individuals with a cleft palate, compared to a cleft lip alone, was tentatively identified. Again, this may be related to the presence of neurological and/or developmental conditions, and thus additional investigation is necessary. A range of other factors may impact upon educational and vocational achievement, including hearing difficulties, school absences, support from teachers and social experiences. Further research is needed to understand these possible deficits in learning, how they may affect individuals within tangible contexts and the type of additional support and intervention which may be needed.

As reported in the Hunt et al. (2005) review, findings as to whether parent-infant interactions are affected in CL/P were mixed. However, in comparison to the review by Hunt and colleagues (2005), the current review cannot conclude whether attachment representations are influenced by more 'severe' cleft types. Nonetheless, parent-infant interactions, as well as family functioning more generally, appear to impact to some degree upon the child's early development and later adjustment. Further investigation of these relationships and the

implementation of appropriate support for families throughout the entire treatment journey is needed (Nelson et al., 2012).

Contradictory findings were identified in the current review in relation to the overall social functioning of individuals affected by CL/P. However, in accordance with the review by Hunt et al., raised levels of specific concerns, including self-consciousness or anxiety in connection with social situations, were observed. Recent research has begun to advance the field via the investigation of contributory factors such as communication difficulties and the influence of perceived teasing and feelings of social acceptance. Thus, although measurement of societal attitudes towards visible difference is intriguing and of value to organisations campaigning for appearance 'equality' (www.changingfaces.org.uk), a focus of individual-level interventions should be to address the more amenable areas of patients' social perceptions and social competence. In addition, further research is needed to address the question of whether romantic relationships and decisions to have children are affected by having CL/P.

Methodological Considerations

The review by Hunt and colleagues (2005) identified several methodological concerns and made a number of recommendations for future research. Utilising the findings of the current review, the degree to which these recommendations have been met will now be examined.

Hunt et al. (2005) reported that more than half of studies included in their review did not use an appropriate control group. The current review distinguished between studies using control groups and those using reference groups, registry databases and/or norms. When taking these factors into account, only 24.8% of the studies included in the current review did not include an appropriate comparison group. Although the figures from the two reviews cannot be directly compared, these calculations do suggest an improvement in the number of studies using an appropriate comparison group during recent years. One additional and significant consideration is that individuals with CL/P may report scores which are lower than controls, but still within the normal range. The inclusion of norms, including cut-off scores to aid interpretation, is therefore strongly recommended. In addition, caution in regard to the generalisation of findings should be taken when considering the results of separate studies which have utilised data extracted from the same cohort of participants.

Hunt and colleagues (2005) also identified a relative lack of longitudinal studies within the available literature (14%). In the current review, only 5% studies measured variables at two

time points or more, albeit during a much shorter time period, suggesting overall that the field has somewhat responded to this need. Nonetheless, the number of studies using a longitudinal approach is clearly comparatively small and must be increased in order for changes in psychological adjustment over time to be explored.

Much like Hunt et al. (2005), the current review was unable to draw conclusions regarding the effect of age on psychological adjustment to CL/P, since results are predominantly confounded by a wide age range spanning several stages of development. Coupled with a small sample size, results are often indecipherable. The challenges of collecting large samples are well acknowledged in this field. However, efforts must be doubled if we are to investigate subgroups, such as gender, ethnicity, cleft type, additional conditions and other demographic variables effectively. Further, Hunt's recommendation of multi-centre studies is just as valid today.

The review by Hunt et al. (2005) reported that a wide variety of measures were in use, making it difficult to compare the results from different studies directly. Unfortunately, this still seems to be the case. A number of recent reviews have also demonstrated the quantity of measures available, coupled with a lack of agreement about what should be measured, how and when (Eckstein, Wu, Akinbiyi, Silver, & Taub, 2011; Klassen et al., 2012; Ranganathan et al., 2015). Psychological adjustment is multifactorial and difficult to capture, making the process of achieving consensus in regard to measurement considerably challenging. Due largely to the centralisation of cleft treatment, progress has recently been made in the UK (Stock, Feragen et al., in press), and elsewhere (Broder, 2014; Semb, 2014). This type of approach could provide a foundation for integrating standardised patient-centred measurement in countries around the world.

Another important methodological consideration identified by Hunt et al. (2005) was discrepancies in the reporting of outcomes depending on the informant. In the current review, many more studies included the patient perspective, and an increase in qualitative work is visible (see also Nelson, 2009). However, several studies still relied solely upon reports from parents, teachers, laypersons and/or health professionals. Collecting multiple perspectives can be informative and is encouraged, but should always seek to include the patients' view.

Finally, Hunt and colleagues (2005) stated that a shift in focus 'towards the strengths rather than the deficits' of the affected individual, was necessary. These suggestions were supported at the time by two other discussion papers (Eiserman, 2001; Strauss, 2001). The current

review has demonstrated that measures of positive adjustment and resilience are beginning to be included in CL/P studies (e.g. Feragen et al., 2009; Chimruang et al., 2011; Gassling et al., 2012; Pisula et al., 2014; Stock et al., 2015; Stock, Feragen et al., in press). This progress should be built upon to provide a more balanced view of adjustment and to allow for the assessment of points of opportunity as well as risk.

A more thorough investigation of conceptual and methodological challenges within the field of CL/P has recently been offered (*manuscript in preparation*). Future studies interested in the psychological adjustment of individuals affected by CL/P and their families are encouraged to take this critique into consideration, in order to advance progress within the field as a whole.

Conclusions

As suggested in a previous and comprehensive literature review (Hunt et al., 2005), the current narrative review found the impact of CL/P on overall psychological adjustment and QoL to be low when examining total scores. However, potential difficulties are found when examining key domains of adjustment in more detail. Despite the volume of work conducted in this area during the last decade, research findings still appear to be largely inconclusive, and marred by similar methodological challenges as noted by Hunt et al. in 2005.

Nonetheless, the present review identified some areas of emerging consensus, and a number of recent improvements in the approaches used are visible. Efforts to collect data from large, representative and longitudinal samples, which are comparable across studies and encompassing of both the patient perspective and measures of resilience, should be doubled.

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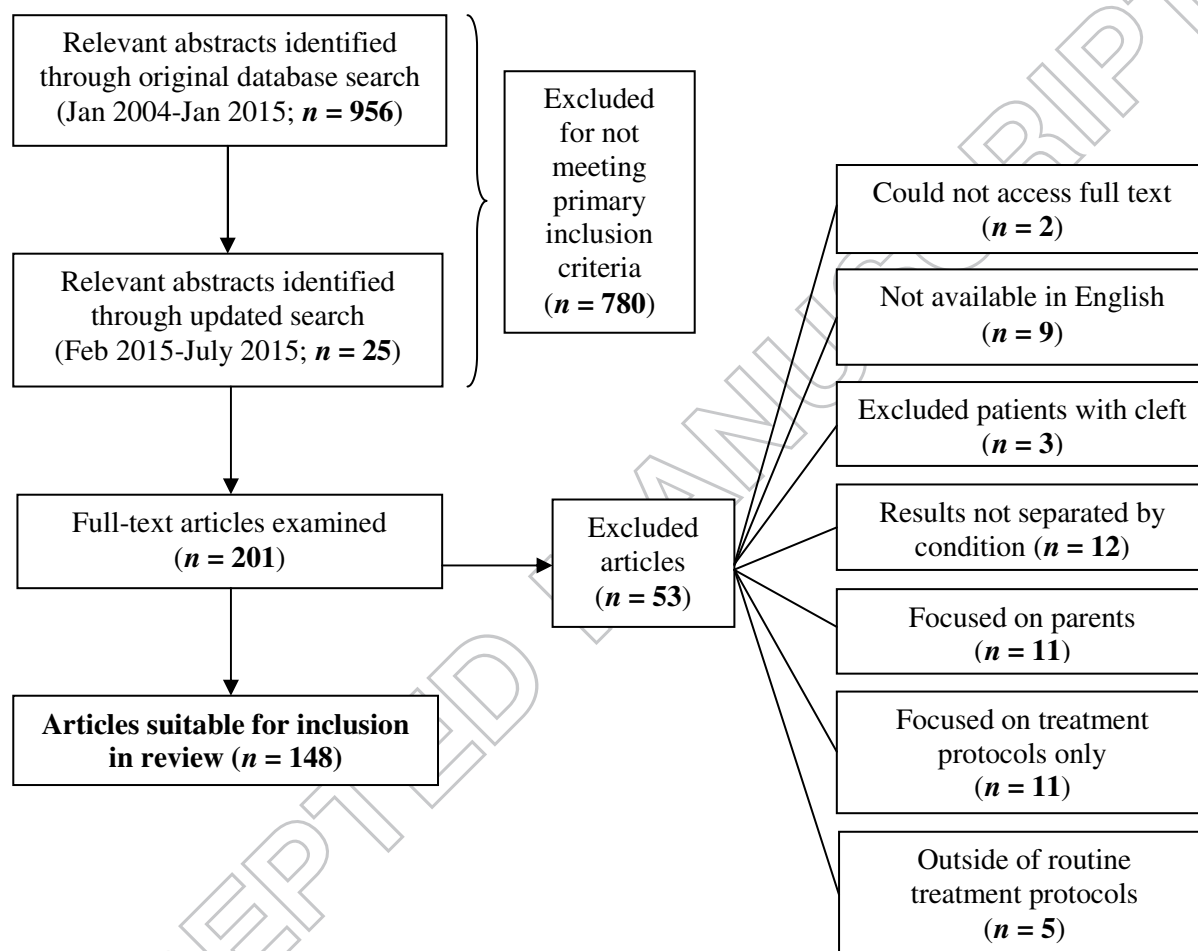
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SUPPLEMENTARY FILE**Figure 1:** Flow chart demonstrating the selection of articles for inclusion in this review.

SUPPLEMENTARY FILE**Table 1:** Search terms used in this review.

Diagnosis	Population	Developmental Trajectory	Behaviour	Emotional Wellbeing	Social Experiences	Appearance and Treatment
Cleft Cleft lip and palate Cleft lip/palate CLP CL/P Craniofac* Visible difference Disfigure* Impair*	Parent* Maternal Famil* Child* Adolesc* Adult	Cognit* Learn* School Achieve* Language Memory Intelligen* IQ Develop* Speech Health	Behaviou* Attention Hyperactivity Internal* External*	Psych* Impact Effect Adjust* Experience Emotion* Affect Stress Depress* Anx* Stigma Cop* Distress Quality of life QoL Self* Resilien*	Social Peer Friend* Intima* Attach*	Treatment Surg* Pathway Satisfaction Function Rating Outcome Body Image Appearance

* denotes unlimited truncation which retrieves all possible suffix variations of a root word.

SUPPLEMENTARY FILE**Table 2:** Categorisation of findings according to five key domains of adjustment.

Developmental Trajectory	Behaviour	Emotional Wellbeing	Social Experiences	Satisfaction with Appearance and Treatment
General health Associated conditions Neurological aspects Cognitive development Language development Educational experiences Employment	Behavioural conduct Internalising/externalising behaviours Inattention/hyperactivity	Psychiatric conditions Emotional functioning Self-concept Self-esteem	Parent-child relationships Social functioning Communication difficulties Social reactions to CL/P Teasing Social acceptance Romantic relationships	General satisfaction with appearance Motivations for surgical intervention Experiences of treatment Satisfaction with aesthetic and functional treatment outcomes

SUPPLEMENTARY FILE**Table 3:** An overview of the articles included in this review.

x = missing data; V = validated; U = unvalidated; B = behaviour; D = developmental trajectory; E = emotional; S = social; G = general adjustment; T = treatment outcome

Author and year	Domain(s) of adjustment	Method	Informant(s)	Sample size	Age range	Comparison group	Recruitment sites	Exclusion criteria	Measures
Gourion et al. (2004)	E	Psychiatric assessments	Professionals	13 patients + 45 parents	21-37 years	42 controls	Single centre, France	x	V and U
Laasonen et al. (2004)	D	Cognitive assessments	Professionals	64 patients	10 years	7 controls	Single centre, Finland	Learning difficulties; difficulties completing the tasks	V
Richman et al. (2004)	D, B	Cognitive assessment	Professionals, parents, teachers	32 patients	7-12 years	Norms	Single centre, USA	No medication	V
Richman et al. (2005)	D	Cognitive assessments	Professionals	48 patients	7-9 years	Norm data	Single centre, USA	Syndromic cleft	V
Scheuerle et al. (2004)	D	Observation	Parents and professionals	56 patients	22-30 months	None	Single centre, USA	x	V
Semb et al. (2005)	S, T	Questionnaires	Patients, parents	93 patients + 82 parents	17 years	None	5 centres, Northern Europe	x	U
Slifer et al. (2004)	E, S	Questionnaires; observation	Patients, parents, professionals	34 patients	8-15 years	34 controls	Single centre, USA	Identified genetic syndrome; diagnosis of mental retardation; significant hearing	V

								impairment; severe speech impairment; full-time special educational services	
Nopoulos et al. (2005)	D, S	Questionnaires; neuroimaging	Patients, professionals	46 patients	18+ years	46 controls	Single centre, USA	Syndromic cleft (van der Woude retained)	V
Sinko et al. (2005)	E, S, T	Questionnaires; medical assessment	Patients, professionals	165 patients	18-30 years	Norm data	Single centre, Austria	Associated conditions	V
Chan et al. (2006)	D	Questionnaires	Parents, teachers, employers	39 parents, 27 teachers, 37 employers	11-16 years	None	Single centre, China	x	V and U
Cheung et al. (2006)	E, S, T	Questionnaires	Patients	9 patients	15-40 years	9 controls	Single centre, China	x	V
Damiano et al. (2006)	D, S, T	Structured interview	Parents	151 mothers	2-12 years	85 mothers of children with club foot	Registry, USA	Syndromic cleft; mothers without full custody	V
Eide et al. (2006)	D	Cognitive assessment	Professionals	611 patients	18 years	Registry data	Registry, Norway	x	V
Frederickson et al. (2006)	S	Observation	Professionals	17 patients	3 years	17 controls	3 centres, USA	Abnormal range of cognitive skills; other congenital anomalies; neurologi	V

								cal impairments; sensorineural hearing loss; syndromic cleft	
Goldsberry et al. (2006)	D	Neuroimaging	Professionals	8 male patients	18+ years	6 controls	Single centre, USA	Left-handed; history of significant medical, neurological or psychiatric illness	N/A
Gussy & Kilpatrick (2006)	D, E, S, T	Questionnaire	Patients	23 patients	12-16 years	Norm data	Single centre, Australia	Significant medical condition or associated syndrome; learning disability; developmental delay; intellectual disability	V
Hunt et al. (2006)	E, S, T	Questionnaires	Patients	160 patients	8-21 years	113 controls	3 centres, UK	Syndromic cleft; learning difficulties; significant medical history	V and U
Landsberger et al. (2006)	T	Questionnaire	Patients	33 patients	10-30+ years	None	Single centre, Germany	x	U
Sandor & Ylikontio (2006)	T	Questionnaires; structured interview	Patients	35 patients	16-59 years	None	Single centre, Canada	x	U
Shriver et	D, E	Neuroimaging;	Patients, professionals	46 male	18-47	46	Single centre,	Syndromic cleft;	V

al. (2006)		cognitive assessments; medical assessment	onals	patients	years	controls	USA	serious medical or psychiatric condition; active substance abuse	
Slifer et al. (2006)	S, T	Observation; emotional response; questionnaires	Patients, professionals	24 patients	7-16 years	25 controls	Single centre, USA	Genetic syndrome with mental retardation; moderate to severe hearing impairment in both ears; severe speech impairment; receipt of full-time special educational services	V
Boes et al. (2007)	D, E, S	Questionnaires; neuroimaging	Patients, parents, professionals	33 male patients	7-12 years	43 male controls	Single centre, USA	Syndromic cleft; medical or neurological disease; history of learning disorder or psychiatric disorder	V
Cheung et al. (2007)	D, E, S	Questionnaires	Patients	94 patients	10-40 years	116 controls	Single centre, China	x	V
Damiano et al. (2007)	T	Questionnaires; structured interview	Parents	104 mothers	2-12 years	Norm data	Registry, USA	Syndromic cleft; mothers without full	V and U

								custody	
Hunt et al. (2007)	E, B, S	Questionnaires; structured interview	Parents	129 parents	8-18 years	96 controls	3 centres, UK	Syndromic cleft; learning disabilities; significant medical history	V and U
Noor & Musa (2007)	E, S, T	Structured interview	Patients and parents	60 patients and their parents	12-17 years	None	Single centre, Malaysia	Syndromic cleft; hearing impairment; neurological impairment; mental retardation	U
Nopoulos et al. (2007)	D	Cognitive assessment; neuroimaging; medical assessment	Professionals	74 patients	7-17 years	74 controls	Single centre, USA	IQ less than 70	V
Oosterkamp et al. (2007)	D, E, S, T	Questionnaires; open-ended questions	Patients	43 BCLP patients	20-36 years	43 controls	Single centre, The Netherlands	Syndromic cleft; associated malformations; psychiatric problems	V and U
Bashir et al. (2008)	D	Retrospective clinical review	Professionals	191 patients	3-8 months	Norm data	Single centre, UK	x	N/A
Chuo et al. (2008)	E, T	Questionnaires; medical assessment	Patients, professionals	145 patients	15-70 years	None	Single centre, UK	Presenting with problems unrelated to cleft	U
Conrad et al. (2008)	D	Cognitive assessment	Professionals	77 patients	7-17 years	89 controls	Single centre, USA	Syndromic cleft	V
Kramer et	E, G	Question	Patients,	147	5-6	None	Single	Primary	V

al. (2008)		naires	parents	patients and their parents	years		centre, Germany	surgical treatment not completed; non-cleft medical treatment 3 months prior to study	
Luoto et al. (2008)	E, S	Questionnaires	Patients	51 patients	11-14 years	82 controls	Single centre, Finland	x	V
Murray et al. (2008)	D, B, S	Longitudinal observation; structured interview	Professionals, parents	94 mothers	0-2 years	96 controls	4 centres, UK	Premature birth; low birth weight; syndromic cleft	V
Persson et al. (2008)	D	Cognitive assessment	Professionals	423 male patients	17-19 years	Registry data	Registry, Sweden	Severe handicap or chronic disease which prevents mandatory military enrolment; other congenital malformation; chromosomal abnormalities; men who have emigrated	V
Scherer et al. (2008)	D	Observation; longitudinal cognitive assessment	Professionals	13 patients	0-2 years	13 controls	Single centre, USA	x	V
Sharp et al. (2008)	T	Semi-structured interview	Parents	75 parents	2-22 years	None	Single region, The	Surgery less than six	U, Content

							Philippines	months prior to interview	Analyses
Berger & Dalton (2009)	D, B, E, S, T	Questionnaires	Patients, parents	134 patients, 143 parents	11-16 years	Norm data	Two centres, UK	Not in full-time secondary education; wider craniofacial condition; cognitive, language or communication difficulties impeding questionnaire completion	V and U
Brand et al. (2009)	D, B, E, S	Questionnaires	Patients, parents	32 patients and their parents	6-16 years	34 controls	Single centre, Switzerland	x	V and U
Brunnegård et al. (2009)	T	Speech assessment; questionnaire	Professionals, laypersons	26 CP patients	9-11 years	10 controls	Single centre, Sweden	Syndromic cleft; other malformations	U
Chetpakkdechit et al. (2009)	S	Semi-structured interviews	Patients	12 patients	24-33 years	None	Single centre, Sweden	x	Grounded Theory
Conrad et al. (2009)	D	Cognitive assessment; speech assessment	Professionals	66 patients	7-17 years	87 controls	Single centre, USA	Syndromic cleft	V
Feragen et al. (2009)	D, E, S	Questionnaires	Patients, parents	268 patients and their parents	10 years	Norm data	Single centre, Norway	Severe developmental problems resulting in incomplete data	V and U

Kramer et al. (2009)	S	Questionnaires	Patients, parents	132 patients and their parents	8-12 years	Norm data	Single centre, Germany	Diagnosed syndrome; medical care unrelated to cleft treatment 3 months prior to study	V
Meyer-Marcotty & Stellzig-Eisenhauer (2009)	T	Questionnaire; 3D facial imaging; assessments of photographs	Professionals, laypersons	30 patient photographs, 10 orthodontists, 10 surgeons, 15 laypersons	18-32 years	None	Single centre, Germany	Syndromic cleft; other congenital facial anomalies; any outstanding facial characteristics (e.g. tattoo, piercing); psychiatric disorders	U
Ruiter et al. (2009)	D, S	Retrospective longitudinal clinical review	Professionals	63 patients	2-3 years and again at 5-6 years	Norm data	Single centre, The Netherlands	Missing data; not speaking Dutch; syndromic cleft; other developmental problem	V and U
Sagheri et al. (2009)	E, G	Structured interview	Parents	61 parents	4-7 years	National survey data	Single centre, Germany	Syndromic cleft	V
Collett, Leroux et al. (2010)	D	Observation; longitudinal cognitive assessment; structured interview	Professionals, parents	57 patients	3 months, 12 months, 2 years, 5 years, 7 years	77 controls	Single centre, USA	Syndromic cleft; additional birth defects; perinatal problems	V

Collett, Stott-Miller et al. (2010)	D	Cognitive assessment; structured interview	Professionals, parents	42 patients	5-7 years	43 controls	Single centre, USA	Syndromic cleft; English not primary language; visual or auditory impairments; history of mental retardation or traumatic brain injury; out of home placement	V
Feragen & Borge (2010)	S	Questionnaires	Patients, parents	661 patients and their parents	10 years, 16 years	Norm data	Single centre, Norway	Severe developmental problems resulting in incomplete data	V and U
Feragen, Kvaalem et al. (2010)	E, S	Questionnaires	Patients	289 patients	16 years	Norm data	Single centre, Norway	Severe developmental problems resulting in incomplete data	V
Mani et al. (2010)	E, S, T	Questionnaires	Patients	86 UCLP patients	20-47 years	Norm data	Single centre, Sweden	Syndromic cleft	V
Meyer-Marcotty, Alpers et al. (2010)	S, T	3D facial imaging; eye-tracking	Laypersons	18 UCLP patient photographs, 18 control photographs, 30 laypersons	17-39 years	20 orthognathic patients with Class III, 20 controls with Class I	Single centre, Germany	No associated malformations or other distinctive facial features; incomplete cleft	U

Meyer-Marcotty, Reuther et al. (2010)	T	3D facial imaging; assessment of photographs	Laypersons	18 patient photographs, 18 control photographs, 30 laypersons	17-39 years	20 orthognathic patients with Class III, 20 controls with Class I	Single centre, Germany	No associated malformations or other distinctive facial features; complete UCLP	U
Murray et al. (2010)	E, B, S	Observation; medical assessment	Parents, patients, teachers, professionals	93 patients	7 years	77 controls	2 centres, UK	Other abnormalities or health problems	V
Nopoulos et al. (2010)	D, B	Questionnaires, neuroimaging	Parents, teachers, professionals	50 male patients	7-17 years	60 controls	Single centre, USA	Syndromic cleft; IQ less than 70; major medical, neurologic or psychiatric illness; learning disability	V
Snyder & Pope (2010)	D, B, S	Questionnaires	Parents	144 parents	2-18 years	Norm data	Single centre, USA	x	V
Van der Plas et al. (2010)	D	Neuroimaging	Professionals	33 male patients	7-17 years	57 controls	Single centre, USA	Syndromic cleft; history of medical or neurologic disease	N/A
Versnel et al. (2010)	E, S, T	Questionnaires	Patients	59 patients	34-45 years	59 patients with facial trauma; 201 controls	Single centre, The Netherlands	Incomplete data; under 18 years of age; mentally retarded; blind; non-Dutch speaking; additional congenital	V and U

								craniofacial condition	
Young et al. (2010)	D	Cognitive assessment	Professionals	43 patients	3-6 years	Norm data	Single centre, China	Syndromic cleft; neurological or additional congenital abnormalities; sensorineural hearing problems; intellectual impairment; developmental delay	V
Yttri et al. (2010)	S	Retrospective clinical review	Professionals	1931 female patients	15+ years	Registry data	Registry, Denmark	Emigration; having children before age 15 years; syndromic cleft; major anomalies	N/A
Augsornwan et al. (2011)	E, T	Questionnaires; semi-structured interview	Patients	33 patients completed questionnaires, 15 patients were interviewed	8-18 years	Norm data	Single centre, Thailand	Other anomalies	V, Content Analysis
Berger & Dalton (2011)	E, S, T	Questionnaires	Patients, parents	91 patients and their mothers	11-16 years	Norm data	2 centres, UK	Other craniofacial condition; cognitive, language or communi	V and U

								ation difficultie s	
Bos & Prahl (2011)	D, E, T	Questionnaire	Patients, parents	122 patients and their parents	8-15 years	Norm data	Single centre, The Netherlands	x	V
Chetpakdeechit et al. (2011)	T	Assessment of photographs	Professionals, laypersons	12 BCLP patient photographs, 25 orthodontists, 20 laypersons	16-19 years	None	Single centre, Sweden	Not completed treatment; mental disorders; other craniofacial defects	U
Chimruang et al. (2011)	E, S, T	Semi-structured interviews, participatory activities	Patients, parents	18 patients, 6 parents	12-17 years	None	Single centre, Thailand	Syndromic cleft; additional disabilities	Content analysis
Demir et al. (2011)	D, E, S, T	Questionnaires; cognitive assessment	Patients, professionals	20 patients	6-16 years	40 controls	Single centre, Turkey	Syndromic cleft; other deformity with cosmetic disfigurement; chronic disease; hearing impairment	V and U
Despars et al. (2011)	S	Semi-structured interview; longitudinal questionnaires	Parents	22 mothers	2-3 months, 1 year	Norm data	Single centre, Switzerland	Syndromic cleft; cleft palate only; parental psychiatric illness or drug abuse; difficulty speaking French	V, Content analysis
Havstam, Laakso,	E, S	Semi-structured	Patients	13	25-34	None	Single centre,	No palatal	Grounded

Lohmander et al. (2011)		interview		patients	years		Sweden	involvement	Theory
Havstam, Laakso & Ringsberg (2011)	S	Longitudinal speech assessment; questionnaires	Patients, parents, professionals	54 patients and their parents	10 years	Norm data	Single centre, Sweden	No palatal involvement; moved away from the region; severe developmental delay; did not attend appointment; lost data	V and U
Havstam, Sandberg et al. (2011)	E, S, T	Semi-structured interview	Patients	13 patients	25-34 years	None	Single centre, Sweden	No palatal involvement	Grounded Theory
Hens et al. (2011)	T	Questionnaire; assessment of photographs	Patients; professionals	30 patients	18-60 years	None	Single centre, Belgium	x	U
Hentges et al. (2011)	D, S	Observation; longitudinal cognitive assessment	Professionals	93 patients	7 years	77 controls	4 centres, UK	No other abnormalities; no other health problems	V
Meyer-Marcotty, Gerdes et al. (2011)	T	Assessment of photographs	Laypersons	30 patient photographs, 20 orthognathic patients, 20 laypersons	17-39 years	20 orthognathic patients, 20 controls	Single centre, Germany	No malformations or other distinctive features	U
Meyer-Marcotty, Kochel et al. (2011)	S	Assessment of photographs; eye-tracking	Laypersons, patients	33 patients, 30 laypersons	23-30 years	30 controls	Single centre, Germany	No other congenital facial anomalies; no other	U

								distinctive facial features	
Munz et al. (2011)	E, S, T	Questionnaires	Patients, parents	27 patients, 30 parents	12-23 years	None	Single centre, USA	Over the age of 25 years; not completed treatment; desire for further treatment	V and U
Pitak-Arnop et al. (2011)	T	Assessment of photographs	Professionals, laypersons	50 patient photographs, 51 professionals, 507 laypersons	15-48 years	None	Single centre, Germany	Syndromic cleft	U
Reekie (2011)	T	Questionnaires	Patients	15 patients of South Asian ethnicity	16-65 years	95 Caucasian patients	Single centre, UK	Syndromic cleft; mental retardation; patients with CP	U
Vogels et al. (2011)	E	Questionnaires	Patients, parents	110 patients	4-12 years	Norm data	Single centre, The Netherlands	x	V and U
Broder et al. (2012)	D, E, T	Questionnaire	Patients	839 patients	7-19 years	Norm data	6 centres, USA	Non-English or non-Spanish speaking	V
Collett et al. (2012)	E, B, S	Questionnaires	Patients	93 patients	5-9 years	124 controls	National survey data, USA	Syndromic cleft; Mendelian-inherited disorder	V
Gassling et al. (2012)	E	Stress test; physiological assessment; questionnaire	Patients, professionals	30 patients	18-40 years	30 controls	Single centre, Germany	x	V and U

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Ha et al. (2012)	D, B, E, S	Questionnaires	Parents	93 patients	6-11 years	100 controls	Single centre, China	CP; other abnormalities or health problems	V
Hall et al. (2012)	E, T	Semi-structured interview and participatory activities	Patients	17 patients	8-17 years	None	Single centre, UK	Learning disability; significant medical history; non-English speaking	Thematic analysis
Montirosso et al. (2012)	S	Questionnaires; observation	Patients, professionals	25 patients	2 months	25 controls	Single centre, Italy	Prematurity; low birth weight; syndromic cleft	V and U
O'Hanlon et al. (2012)	E, S	Questionnaires, open-ended questions	Patients, parents	27 parents with cleft	M = 45 years	27 parents without cleft	Single centre, UK	Another immediate family member with cleft	V and U
Papamantou et al. (2012)	T	Assessment of photographs	Laypersons, professionals	12 UCLP patient photographs, 12 laypersons, 12 orthodontists, 12 surgeons	17-27 years	None	Single centre, Greece	Professionals with less than 10 years' experience	U
Van Lierde et al. (2012)	S, T	Questionnaire	Patients, parents	43 UCLP	10-17 years	43 controls	Single centre, Belgium	Syndromic cleft; cognitive deficiency; neuromotor dysfunction; residual hard palate fistula; hearing thresholds below 20db in the	U

								poorer ear	
Wehby et al. (2012)	D, B, E	Structured interview	Parents	104 mothers	2-12 years	Norm data	Registry data, USA	Syndromic cleft	V
Young et al. (2012)	D	Cognitive assessment	Professionals	86 patients	3-6 years	100 controls	Single centre, Singapore	Failed hearing screening	V
Abd-Elseyed et al. (2013)	E	Participatory activities	Patients, professionals	171 patients	5-17 years	None	Medical mission, 22 countries (majority USA and Canada)	x	U
Alansari et al. (2013)	E, S, T	Semi-structured interview	Patients	11 UCLP and BCLP patients	19-54 years	None	Non-profit organisation, 3 cities in Canada	Syndromic cleft; incomplete cleft; under 18 years of age; non-English speaking	Grounded Theory
Dogan et al. (2013)	E	Questionnaires	Patients	17 patients	12 years	15 controls	Single centre, Turkey	x	V
Eslami et al. (2013)	E	Questionnaire	Patients	50 patients	8-12 years	Norm data	Single centre, Iran	x	V
Foo et al. (2013)	T	Assessment of photographs	Patients, laypersons, professionals	80 patient photographs, 2 patients, 2 laypersons, 5 professionals	18-64 years	None	Single centre, Australia	Syndromic cleft; under 18 years of age; not completed treatment; not treated at a single centre	U
Gkantidis et al. (2013)	D, S, T	Assessment of photographs	Patients, parents, laypersons, professionals	12 UCLP patients, 12 parents, 12 laypersons, 12 professionals	17-27 years	None	Single centre, Greece	Professionals with less than 10 years' experience	U

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Hall et al. (2013)	E, S, T	Semi-structured interview; participatory activities	Patients	17 patients	8-17 years	None	Single centre, UK	Learning disability; significant medical history; not fluent in English	Narrative
Mani et al. (2013)	E, T	Questionnaires; assessment of photographs	Patients, professionals	86 UCLP patients	20-47 years	Norm data	Single centre, Sweden	Syndromic cleft; incomplete cleft; major mental or physical incapacity; emigrated	V and U
Millar et al. (2013)	D, B, E, T	3D imaging; questionnaire	Patients, parents, professionals	94 patients	10 years	68 controls	Single centre, UK	English is not first language	V
Nkenke et al. (2013)	T	Questionnaire	Patients	362 patients	6-18 years	None	Single centre, Germany	x	U
Tannure et al. (2013)	G	Structured interview	Patients	35 patients	5-12 years	35 controls	Single centre, Brazil	Clinically or genetically diagnosed syndrome	V
Tiemens et al. (2013)	S, T	Semi-structured interviews	Patients	7 female patients	15-20 years	None	Single centre, Canada	Not scheduled for orthognathic surgery	IPA
Tyler et al. (2013)	S, T	Questionnaires	Parents	271 mothers, 8 fathers	4-9 years	Norm data	3 centres, USA	No longer living with biological parent; developmental delay or mental retardation	V and U

Van der Plas et al. (2013)	D, E, S	Questionnaires; educational records; eye-tracking; emotional response; physiological assessment	Patients, professionals	20 male patients	13-25 years	20 controls	Single centre, USA	Syndromic cleft; history of severe hearing loss requiring a hearing aid; IQ lower than 70; history of epilepsy, brain trauma or brain tumour	V
Watterson et al. (2013)	E, T	Speech assessment	Professionals, laypersons	7 patients, 44 laypersons, 1 professional	8-11 years	3 siblings	Single centre, UK	x	U
Yunusa & Obembe (2013)	E	Questionnaires	Patients	200 patients	20-39 years	Norm data	Single centre, Nigeria	Presence of overt or past psychiatric ailment; poor level of literacy	V and U
Adamson et al. (2014)	D	Neuroimaging	Professionals	26 patients	6-14 years	26 controls	Single centre, Australia	Syndromic cleft; complex medical condition	N/A
Broder., Wilson-Genderson & Sischo (2014)	E, T	Questionnaires	Patients	1,200 patients	7-18 years	Norm data	6 centres, USA	Non-English or non-Spanish speaking	V
Broder., Wilson-Genderson, Sischo et al. (2014)	T, G	Questionnaire	Patients	1,200 patients	7-19 years	Norm data	6 centres, USA	Non-English or non-Spanish speaking	V
Burnell et al. (2014)	D	Retrospective clinical	Professionals	81 patients	2+ year	None	Single centre,	Less than 2 years' follow-up	N/A

		review			s		Canada	data	
Byrne et al. (2014)	T	Questionnaire; open-ended questions; assessment of photographs	Patients, professionals	35 patients, 45 professionals	14-53 years	None	Single centre, Ireland	Not requiring secondary rhinoplasty	V and U
Collett et al. (2014)	D	Educational records	Professionals	256 patients	7-19 years	387 unaffected siblings	Registry, USA	Syndromic cleft; non-biological siblings	N/A
Conrad et al. (2014)	D, B	Cognitive assessment	Professionals	80 patients	7-26 years	62 controls	Single centre, USA	Head trauma; major medical disorder; English not dominant language; learning disabilities; exceptional achievement	V
Eichenberger et al. (2014)	T	Assessment of photographs	Laypersons, professionals	20 patients	M = 20.5 years	10 patients with Class I occlusion	Single centre, Switzerland	Syndromic cleft; other congenital anomalies; outstanding facial characteristics; incomplete records; not completed recommended treatment	U
Feragen, Stock &	D	Questionnaires;	Patients	754 patients,	10 year	Norm data	Single centre,	Severe developm	V

Rumsey (2014)		retrospective clinical review		169 with psychology data	s		Norway	ental problems resulting in incomplete data	
Feragen & Stock (2014)	D, B, E, S	Questionnaires; retrospective clinical review	Patients	205 patients	10 years	Norm data	Single centre, Norway	Severe developmental problems resulting in incomplete data	V
Gassling, Christoph et al. (2014)	S	Observation	Professionals	15 patients	8-12 years	20 controls, 20 migraine patients	Single centre, Germany	Syndromic cleft	U
Gassling, Kessler et al. (2014)	E, S	Questionnaire; emotional response	Patients, professionals	25 patients	18-40 years	25 controls	Single centre, Germany	Psychiatric comorbidity	V
Pisula et al. (2014)	E, G	Questionnaires	Patients	48 patients	16-23 years	48 controls	Single centre, Poland	Under 16 years of age; syndromic cleft; intellectual or learning disability	V
Smith et al. (2014)	D, E, G	Medical assessment; longitudinal; cognitive assessment	Professionals	33 patients	3 years	Norm data	Single centre, Canada	Medically unstable; required previous medical interventions	V
Wehby, Collett et al. (2014)	D	Educational records	Professionals	588 patients	7-16 years	Registry	Registry data, USA	Syndromic cleft	N/A
Wehby, Nyarko et al. (2014)	E	Questionnaires	Patients	182 parents with cleft	27-30 years	Reg	National survey, Brazil	No children at time of enrolment	V
Crerand	S	Question	Parents	1,200	7-18	Norm	6 centres,	Non-	V and

et al. (2015)		naires		parents	years	data	USA	English or Spanish speaking; inability to read at second grade level; craniofacial syndrome; complex medical condition	U
Feragen et al. (2015)	D, B, E, S	Questionnaires	Patients, parents	857 patients, 304 caregivers	16 years	Norm data	Single centre, Norway	Severe developmental problems resulting in incomplete data	V and U
Ferrari & Mauro (2015)	T	Assessment of photographs	Professionals, laypersons	23 BCLP patient photographs, 10 cleft professionals, 10 non-cleft professionals, 5 laypersons	19-41 years	None	Single centre, Brazil	Under 18 years of age; non-White Brazilian patients; syndromic cleft; not completed all treatment; partly treated at another centre	U
Gkantadis et al. (2015)	D, S, T	Structured interview	Patients, parents	33 patients, 30 parents	9-33 years	None	Single centre, Greece	Under 9 years of age; syndromic cleft; other congenital anomalies with associated malformations; moderate	U

								to severe mental retardation	
Hamlet & Harcourt (2015)	E, S, T	Semi-structured interview	Patients	6 patients	57-82 years	None	Advertisements, UK	Under the age of 55 years	IPA
Klinto et al. (2015)	D	Observation; cognitive assessment	Professionals	29 UCLP patients	5 years	20 controls	2 centres, Sweden	Non-Swedish speaking; syndromic cleft; additional malformations	V
Knight et al. (2015)	D	Questionnaires	Parents	112 mothers	5-12 years	138 controls	Survey data, USA	Syndromic cleft; unable to speak or read English or Spanish; child born outside of the state	V
Krikken et al. (2015)	E	Questionnaires	Patients, parents	102 patients and their parents	4-21 years	Norm data	Single centre, The Netherlands	x	V
Lee et al. (2015)	D	Cognitive assessment; speech assessment	Professionals	15 patients	6-8 years	15 controls	Single centre, Malaysia	English not dominant language; syndromic cleft	V and U
Lima et al. (2015)	E	Questionnaire	Patients	61 patients	7-17 years	61 controls	Brazil	x	V
Lorot-Marchand et al. (2015)	D, S, T	Questionnaires	Patients	55 patients	12-29 years	None	3 centres, France	x	U
Pausch et al. (2015)	S	Assessment of photographs	Medical and non-medical laypersons	50 patient photographs; 273	15-48 years	None	Single centre, Germany	Syndromic cleft; incomplete	U

			ns	medical and non-medical laypersons				treatment and rehabilitation	
Petrackova et al. (2015)	D, E, T	Questionnaires; cognitive assessment	Professionals, parents	32 patients and their parents	3-7 years	Norm data	Single centre, Czech Republic	No cleft of the lip	V
Smillie (2015)	T	Retrospective clinical review	Professionals	74 patients with cleft	16-28 years	None	Single centre, UK	x	N/A
Stock, Feragen et al. (2015)	D, E, S, T	Semi-structured interview	Patients	52 patients	22-77 years	None	Advertisements, UK	Under 18 years of age	Thematic analysis
Stock & Rumsey (2015)	E, S, T	Semi-structured interview	Patients	24 patients	28-70 years	None	Advertisements, UK	Under 18 years of age; no children	Thematic analysis
Tierney et al. (2015)	D, B, E, S	Semi-structured interview; participatory activities	Parents, patients	37 parents, 22 patients	0-11 years	None	2 centres, UK	Syndromic cleft; no experience of OME; non-English speaking; psychosocial difficulties as reported by the cleft team	Framework analysis
Aravena et al. (in press)	D, E, S	Questionnaire	Patients	48 patients	8-15 years	96 controls	3 cities, Chile	Disabling medical condition; syndromic cleft; mental disorders	V
Feragen Særvold et al. (in press)	D, E, S	Questionnaires; speech assessment	Patients, parents, professionals	170 patients, 170 caregivers	10 years	Norm data	Single centre, Norway	Severe developmental problems resulting in	V and U

								incomplete data	
Feragen & Stock (in press)	D, B, E, S, T	Questionnaires	Patients, parents	845 patients, 722 caregivers	10 years	Norm data	Single centre, Norway	Severe developmental problems resulting in incomplete data	V and U
Kortelainen et al. (in press)	G	Questionnaire	Patients	26 patients	11-14 years	71 controls	Single centre, Finland	x	V
Stock et al. (in press)	E, S, T	Semi-structured interview	Patients	52 patients	22-77 years	None	Advertisements, UK	Under 18 years of age	Thematic analysis
Van Schijndel et al. (in press)	S	Assessment of photographs	Laypersons	18 patient photographs, 40 laypersons	16-28 years	None	Single centre, The Netherlands	Syndromic cleft	U