

The psychosocial effects of cleft lip and palate: a systematic review

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SUMMARY This systematic review examined the published scientific research on the psychosocial impact of cleft lip and palate (CLP) among children and adults. The primary objective of the review was to determine whether having CLP places an individual at greater risk of psychosocial problems. Studies that examined the psychosocial functioning of children and adults with repaired non-syndromal CLP were suitable for inclusion. The following sources were searched: Medline (January 1966–December 2003), CINAHL (January 1982–December 2003), Web of Science (January 1981–December 2003), PsycINFO (January 1887–December 2003), the reference section of relevant articles, and hand searches of relevant journals. There were 652 abstracts initially identified through database and other searches. On closer examination of these, only 117 appeared to meet the inclusion criteria. The full text of these papers was examined, with only 64 articles finally identified as suitable for inclusion in the review. Thirty of the 64 studies included a control group. The studies were longitudinal, cross-sectional, or retrospective in nature.

Overall, the majority of children and adults with CLP do not appear to experience major psychosocial problems, although some specific problems may arise. For example, difficulties have been reported in relation to behavioural problems, satisfaction with facial appearance, depression, and anxiety. A few differences between cleft types have been found in relation to self-concept, satisfaction with facial appearance, depression, attachment, learning problems, and interpersonal relationships. With a few exceptions, the age of the individual with CLP does not appear to influence the occurrence or severity of psychosocial problems. However, the studies lack the uniformity and consistency required to adequately summarize the psychosocial problems resulting from CLP.

Introduction

Research has shown that attractive children are seen by others as brighter, as having more positive social behaviour and receive more positive treatment than their less attractive counterparts (Dion *et al.*, 1972). Many children with cleft lip and palate (CLP) may have a less attractive facial appearance or speech than their peers. A high incidence of teasing over facial appearance is reported among those with CLP (Bernstein and Kapp, 1981; Heller *et al.*, 1981; Noar, 1991, 1992; Turner *et al.*, 1997). The general assumption that follows is that children with CLP must experience some kind of psychosocial distress as a result of their condition.

A number of literature reviews have been conducted describing the psychological status of individuals with CLP (McWilliams, 1982; Richman and Eliason, 1982; Madison, 1986; Eliason, 1991; Tobiasen and Hiebert, 1993; Turner *et al.*, 1998; Endriga and Kapp-Simon, 1999; Thompson and Kent, 2001; Lockhart, 2003). Overall, these reviews appear to conclude that children and adults with clefts do not suffer from any significant psychopathology. However, in all of the reviews, reports of difficulties in particular areas of functioning have been reported. For example, Endriga and

Kapp-Simon (1999) summarized that significantly more children than would be expected demonstrate cognitive, behavioural and emotional difficulties that are of clinical concern. Thompson and Kent (2001) pointed out heightened levels of depression and anxiety among those with facial disfigurement, while Richman and Eliason (1982) noted evidence to indicate behavioural inhibition and concern regarding appearance. Social isolation, speech and language difficulties, specific and global learning difficulties and poor self-image were identified as problem areas in a review by Lockhart (2003). These reviews also point out the methodological weaknesses of previous research (Turner *et al.*, 1998; Thompson and Kent, 2001), including the lack of longitudinal studies and comparison groups, as well as the variability in measurement methods. All of these factors make it difficult to draw definite conclusions.

In the light of the conflicting evidence presented in previous literature reviews, this investigation set out to systematically review the literature for scientific evidence to support or refute the assumption that individuals with CLP experience greater psychosocial problems than those who do not have CLP. It was anticipated that by systematically

reviewing the literature, difficulties in drawing conclusions from previous reviews may be reduced.

Objectives

The overall aim was to evaluate the scientific evidence linking CLP with an increased risk of psychosocial problems. The review addressed a number of specific questions:

1. Are children and adults with CLP at increased risk of impaired psychosocial functioning?
2. What, if any, type of psychosocial impairment do children and adults with CLP develop?
3. Is there a relationship between cleft type and the prevalence and severity of psychosocial impairment?
4. Are children with CLP more vulnerable to psychosocial problems at particular stages of development?

Materials and methods

Inclusion criteria

Randomized controlled trials, longitudinal studies with and without controls, cross-sectional investigations with and without controls, and retrospective studies with and without controls, were considered suitable for inclusion. Only studies that focused on children and adults with repaired non-syndromal CLP were included. No limitation regarding the age of the subjects was imposed. Both self-reports and the reports of others regarding children and adults with CLP were considered suitable. All methods of measurement were considered suitable, including questionnaires (validated and unvalidated), interviews (unstructured, semi-structured and structured), observation and clinical assessment. The searches were not restricted to English language journals.

Exclusion criteria

Literature reviews were excluded from this systematic review. However, previous reviews were identified in order to establish their findings and, therefore, provide a background to previous research in this area. Studies that involved participants with other anomalies or medical conditions and where the results of the study were not reported specifically in relation to those with CLP were excluded. Case studies and dissertation abstracts were also excluded.

For the purpose of this review, psychosocial problems were defined as involving:

1. Psychological functioning, personality and adjustment.
2. Self-concept (including self-esteem and self-confidence).
3. Body image and satisfaction with facial appearance.
4. Satisfaction with speech.
5. Behavioural problems.
6. Social functioning.
7. Anxiety and depression.
8. Attachment, development and learning.

Search strategy for the identification of studies

Medline (January 1966–December 2003), CINAHL (January 1982–December 2003), PsycINFO (January 1887–December 2003), and Web of Science (January 1981–December 2003) were searched using a number of keywords (see Appendix). The Boolean operator ‘AND’ was used to combine and narrow the searches. All variations of the term cleft lip and palate (i.e. CLP, cleft lip and palate, cleft lip, cleft palate) were searched in combination with all other search terms detailed in the Appendix. No language limitations were imposed.

The reference section of the articles identified as suitable for inclusion through Medline, CINAHL, Web of Science and PsycINFO searches were examined to identify other relevant studies. A number of relevant journals were hand searched (1992–2003), including the *Cleft Palate-Craniofacial Journal* (formerly the *Cleft Palate Journal*), *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery* (formerly *Scandinavian Journal of Plastic and Reconstructive Surgery*), and the *British Journal of Plastic Surgery*.

One author (OH) conducted the literature search. In total, 652 abstracts were identified from this search of the databases. A second author (DB) was given a random sample of 50 of these abstracts to determine their suitability or otherwise for inclusion in the review. At this point, there was 96 per cent agreement between the two reviewers. The full text article was obtained of two abstracts for which there was disagreement. The reviewers re-evaluated these studies in the light of the agreed selection criteria.

From the 652 abstracts identified, only 113 were considered suitable for inclusion in the review based on the inclusion and exclusion criteria (Figure 1). The full texts of these papers were obtained, including eight foreign language papers for which English translations were obtained. Four other articles were identified through hand searching relevant journals. This resulted in a total of 117 full text papers being examined. The authors were unable to obtain the full text for one of the studies (Marcusson, 2001). However, from the detailed abstract available, the study appeared to meet the inclusion criteria and was, therefore, included in the review.

Quality assessment

A proforma was prepared to identify the following features of each study: type of participants (i.e. children, adolescents or adults), age range, sample size, method of measurement, inclusion of control group, type of psychosocial problems investigated, whether the self-report of the person with CLP or reports of others were used, and the major findings. This evaluation was used to determine if the study was suitable for inclusion in the review. One author (OH) completed a proforma for each study. To assess reliability, another author (DB) completed proformas for 10 randomly selected articles from the 117 studies identified as suitable. At this stage,

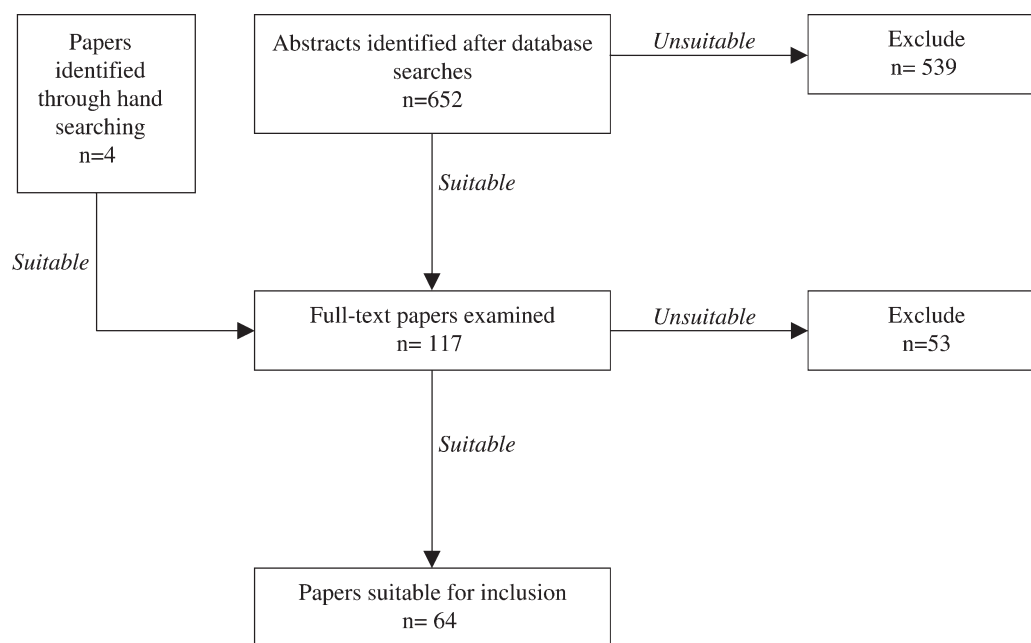


Figure 1 Flow chart demonstrating the selection of studies suitable for inclusion in the review.

complete agreement existed between the two authors. On examination of the full text, only 64 papers were considered to meet the full inclusion criteria (Figure 1).

Twenty-five cross-sectional studies that included a control group were identified through the literature search (Table 1). The sample sizes ranged from 23 to 431 subjects with clefts. The age range of the subjects was 3 months to 69 years. A variety of measures was employed, including validated and unvalidated questionnaires and semi-structured interviews.

Twenty-six cross-sectional studies that did not include control groups were identified (Table 1). Sample sizes ranged from 28 to 495 subjects with clefts, ranging in age from birth to 53 years. The study by Tobiasen and Hiebert (1984) included the responses of 512 parents who served as a standardization sample in the development of a questionnaire (normative data). Again, a variety of measures was employed, including validated and unvalidated questionnaires and semi-structured interviews.

Among the 13 remaining studies (Table 1) included in the review, five were longitudinal in design and included a control group (Hoeksma *et al.*, 1996; Jocelyn *et al.*, 1996; Speltz *et al.*, 1997; Maris *et al.*, 2000; Coy *et al.*, 2002), and two were longitudinal but did not include a control group (Heller *et al.*, 1985; Richman, 1997), although one of these studies included two groups of children with either cardiovascular or hearing disorders as comparison (Heller *et al.*, 1985).

Two studies were retrospective in nature (Bernstein and Kapp, 1981; Broder *et al.*, 1998) and four presented data using more than one research design, either longitudinal and cross-sectional or cross-sectional and retrospective (Starr *et al.*, 1977; Tyl *et al.*, 1990; Richman and Millard, 1997;

Kapp-Simon and Krueckeberg, 2000). Among these studies sample sizes ranged from 32 to 106 subjects with CLP, with participants varying in age from 3 months to 18 years. A variety of measures was employed, including validated and unvalidated questionnaires, semi-structured interviews and observation.

Among the 64 studies (with or without control groups), 13 included reports by others, usually a parent or teacher, as well as a self-report by the individual with CLP (Table 1). In a number of studies ($n = 21$), only the reports of others were obtained, such as parents, teachers, speech and language therapists, or psychologists (Table 1).

Some of the studies examined a single psychological or social construct, while others addressed a range of psychosocial issues (Table 1). Due to the heterogeneity of the study methodologies included in this review it was not possible to apply the traditional methods of a systematic review. A meta-analysis is only suitable if there is sufficient similarity in the populations studied and the measurements used. This was not the case with the studies identified in this review. Therefore, a narrative approach was taken to report the findings of the included studies.

Results

Are children and adults with CLP at increased risk of impaired psychosocial functioning?

At a superficial level, the literature suggests that an individual's psychosocial well-being is not greatly affected by having a CLP (Wiris and Plotkin, 1971; Richman, 1983; Heller *et al.*, 1985; Geier and Wittstock, 1986;

Table 1 Details of the studies included in the review.

Author and year	Focus of the study	Sample size*	Method	Validated (V)/unvalidated (U)	Indication of psychosocial problems	Self-report or other
<i>Longitudinal studies with controls (n = 5)</i>						
Coy <i>et al.</i> (2002)	Facial appearance, attachment	126	Observation	V	No	Other
Hoeksma <i>et al.</i> (1996)	Attachment	100	Observation	V	No	Other
Jocelyn <i>et al.</i> (1996)	Cognition, communication	48	Observation	V	Yes	Other
Speltz <i>et al.</i> (1997)	Attachment	115	Observation and questionnaires	V and U	No	Other
Maris <i>et al.</i> (2000)	Attachment	107	Observation	V	No	Other
<i>Longitudinal studies without controls (n = 2)</i>						
Heller <i>et al.</i> (1985)	Behavioural problems	48†	Interview and questionnaires	V and U	No	Both
Richman (1997)	Behavioural problems	65	Questionnaires	V and U	Yes	Other
<i>Cross-sectional studies with controls (n = 25)</i>						
Berk <i>et al.</i> (2001)	Social anxiety	170	Questionnaires	V	Yes	Self-report
Brantley and Clifford (1979a)	Cognition, self-concept, body image	151†	Questionnaires	V and U	No	Self-report
Brantley and Clifford (1979b)	Locus of control, field dependence	105	Questionnaires	V	Yes	Both
Broder and Strauss (1989)	Self-concept	58	Questionnaire	V	Yes	Self-report
Broder <i>et al.</i> (1994)	Satisfaction with facial appearance, adjustment	559	Interview	U	Yes	Self-report
Chapman <i>et al.</i> (1998)	Conversational skills	40	Observation	V	Yes	Other
Endriga and Speltz (1997)	Mother–infant interaction	116	Observation	V	No	Other
Kapp (1979)	Self-concept	68	Questionnaire	V	Yes	Self-report
Kapp-Simon (1986)	Self-concept	222	Questionnaire	V	Yes	Self-report
Kasuya <i>et al.</i> (2000)	Perception of family	160	Kinetic family drawings	V	Yes	Self-report
Marcusson (2001)	Quality of life, satisfaction with facial appearance, psychosocial distress	68‡	Questionnaire	‡	Yes	Self-report
Marcusson <i>et al.</i> (2002)	Satisfaction with facial appearance, quality of life, body image, somatization, depression	134	Questionnaires	V and U	Yes	Self-report
Persson <i>et al.</i> (2002)	Self-concept, introversion	86	Questionnaires	V	No	Self-report
Peter and Chinsky (1974a)	Marriage	595	Questionnaire	U	Yes	Self-report
Peter and Chinsky (1974b)	Educational achievement	594	Questionnaire	U	Yes	Self-report
Peter and Chinsky (1975)	Social integration	594	Questionnaire	U	Yes	Self-report
Peter <i>et al.</i> (1975)	Vocational and economic aspects	594	Questionnaire	U	Yes	Self-report
Ramstad <i>et al.</i> (1995a)	Social issues (education, employment, marriage)	1557	Questionnaire	U	No	Self-report
Ramstad <i>et al.</i> (1995b)	Social and psychological adjustment	1557	Questionnaire	U	Yes	Self-report
Richman (1976)	Behavioural problems, achievement	88	Questionnaire	V	Yes	Other
Richman (1983)	Social problems, satisfaction with speech and facial appearance, personality adjustment.	60	Interviews and questionnaires	U and V	Yes	Self-report
Slifer <i>et al.</i> (2003)	Behavioural problems, self-concept, social support, quality of life, social interaction, social skills, satisfaction with appearance	68	Questionnaires	V	Yes	Both

Table 1 Continued

Author and year	Focus of the study	Sample size*	Method	Validated (V)/ unvalidated (U)	Indication of psychosocial problems	Self-report or other
Speltz <i>et al.</i> (1993)	Behavioural problems, self-concept	33	Questionnaires	V	No	Both
Starr (1978)	Self-esteem, behaviour problems	120	Questionnaires	V	No	Self-report
Wirls and Plotkin (1971)	Personality factors	132	IQ test, projective personality tests	V and U	No	Self-report
<i>Cross-sectional studies without controls (n = 26)</i>						
Bjornsson and Agustsdottir (1987)	Social characteristics, attitude towards consequences of clefts, appearance and treatment	63	Questionnaire	U	No	Self-report
Bressman <i>et al.</i> (1999)	Quality of life	156	Questionnaires	V	No	Both
Broder <i>et al.</i> (1992)	Satisfaction with appearance and speech	495	Interview	U	No	Both
Clifford (1969)	Severity of condition according to parent	60	Interview and questionnaires	V and U	Yes	Other
Clifford <i>et al.</i> (1972)	Accomplishment, satisfaction with appearance, satisfaction with treatment	98	Interview and questionnaires	U	No	Self-report
Cochrane and Slade (1999)	Appraisal, coping	51	Questionnaires	V and U	Yes	Self-report
Geier and Wittstock (1986)	Social relationships	51	Questionnaire	V	No	Self-report
Harper and Richman (1978)	Personality	52	Questionnaire	V	Yes	Self-report
Heller <i>et al.</i> (1981)	Social and family interactions, work and educational performance	96	Interview	U	Yes	Self-report
King <i>et al.</i> (1993)	Self-evaluation, self-concept	17†	Questionnaires	V	No	Self-report
Leonard <i>et al.</i> (1991)	Self-concept	105	Questionnaire	V	No	Self-report
McWilliams and Musgrave (1972)	Psychological impact of articulation disorders	170	Interview and questionnaires	V and U	No	Other
McWilliams and Paradise (1973)	Educational, occupational and marital status	115	Interview	U	Yes	Other
Millard and Richman (2001)	Adjustment and learning characteristics, behavioural problems, depression and anxiety	65	Questionnaires	V and U	Yes	Both
Neiman and Savage (1997)	Development	186	Questionnaires	V	Yes	Other
Noar (1991)	Treatment aspects, facial appearance and speech, social and emotional aspects of clefts, success of specialists involved	28	Questionnaire	U	Yes	Both
Noar (1992)	Treatment, facial appearance, speech, social and emotional aspects of cleft, success of specialists	109	Questionnaire	U	Yes	Other
Richman (1978)	Behavioural problems	136	Questionnaire	V	Yes	Other
Richman <i>et al.</i> (1985)	Appearance, behavioural problems and personality adjustment	36	Questionnaires	V and U	Yes	Both
Schneiderman and Auer (1984)	Behavioural problems	58	Questionnaire	V	Yes	Other
Starr (1980a)	Self-esteem, behavioural problems, attitude towards clefting	94	Questionnaires	V	No	Self-report
Starr (1980b)	Facial attractiveness, behavioural problems	49	Questionnaires	V and U	No	Both
Starr (1982)	Physical attractiveness and self-esteem	67	Questionnaires	V	No	Self-report
Thomas <i>et al.</i> (1997)	Satisfaction with facial appearance	111	Questionnaires	V	Yes	Both

Table 1 Continued

Author and year	Focus of the study	Sample size*	Method	Validated (V)/ unvalidated (U)	Indication of psychosocial problems	Self-report or other
Tobiasen and Hiebert (1984)	Conduct problems	41	Questionnaire	V	No	Other
Turner <i>et al.</i> (1997)	Psychological, functioning, satisfaction with treatment	112	Interview and questionnaire	U	Yes	Both
<i>Retrospective studies without controls (n = 2)</i>						
Bernstein and Kapp (1981)	Body image, psychosocial problems	60	Interview and observation	V	Yes	Both
Broder <i>et al.</i> (1998)	Learning disability, school achievement, grade retention.	168	IQ test, school reports	V	Yes	Other
<i>More than one research design (n = 4)</i>						
Kapp-Simon and Krueckeberg (2000)	Mental development	180§ 85¶	Observation	V	Yes	Other
Richman and Millard (1997)	Behavioural problems, achievement	44	Questionnaires	V and U	Yes	Other
Starr <i>et al.</i> (1977)	Mental, motor and social behaviour	75§ 28¶	Observation	V	No	Other
Tyl <i>et al.</i> (1990)	Stigmatization self-awareness, parent/child relationships	109	Interview and questionnaires	U and V	Yes	Both

*Relates to the number of individuals with cleft lip and palate and controls where appropriate.

†Excludes subgroups of subjects with other conditions.

‡Information not available from abstract.

§Cross-sectional data.

¶Longitudinal data.

Bjornsson and Agustsdottir, 1987; Bressman *et al.*, 1999). The literature has tended to report the psychosocial functioning of CLP children in a general way. This has often disguised the specific problems that these children have in relation to adjustment, behavioural problems, self-concept, self-esteem, self-confidence, body image, satisfaction with facial appearance, satisfaction with speech, social life, anxiety and depression, and learning problems. These issues are discussed in greater detail below.

What type of psychosocial impairment do children and adults with CLP develop?

Psychological functioning, personality and adjustment.

While overall psychosocial functioning appears to be good among children and adults with CLP, it has been reported that adults may be at risk of impaired psychosocial functioning as a result of CLP, but that this impairment is related to concerns with facial appearance and speech (Ramstad *et al.*, 1995b). A small number of studies go further to suggest that the burdens of adjustment for those with CLP persist into adulthood (Bernstein and Kapp, 1981), with some individuals showing a high level of distress as a result of CLP (Cochrane and Slade, 1999).

Children with CLP have been described as more external in locus of control (i.e. believe their life is determined by

fate, luck or others) and significantly more field dependent (i.e. sensitive to the social environment) than non-cleft controls (Brantley and Clifford, 1979b). Children with CLP also tend to have higher levels of hostility, negative self-worth, a negative outlook and greater dependence compared with the general population (Tyl *et al.*, 1990).

Other studies have addressed children with CLP from the point of view of the child's perception of their parents. Children with CLP tend to see their parents as having more negative feelings and worrying more (Brantley and Clifford, 1979a; Tyl *et al.*, 1990; Kasuya *et al.*, 2000), and adolescents express a lower degree of perceived parental acceptance than controls (Brantley and Clifford, 1979b).

Self-concept problems.

Self-concept appears to be good among children with CLP (Kapp, 1979; Leonard *et al.*, 1991; Persson *et al.*, 2002), and in some cases is better than in control children (Brantley and Clifford, 1979a). Interestingly, 15 per cent of the children in a study by Leonard *et al.* (1991) had abnormally high self-concept scores. However, when specific components of self-concept are examined some problems emerge. Children with CLP may have lower personal and social self-concept scores than controls (Broder and Strauss, 1989) and are

more 'at risk' of having a poor self-concept compared with controls (Kapp-Simon, 1986).

While self-esteem is considered to be generally good among children with CLP (Starr, 1978; Brantley and Clifford, 1979a), the construct of self-esteem has been linked to the child's opinion of his or her facial appearance. Children who are more accepting of their cleft tend to have higher self-esteem (Starr, 1978), and the more physically attractive a person rates themselves the higher that person's self-esteem tends to be (Starr, 1982). Chinese adults with CLP have been found to have lower self-esteem than control subjects and siblings (Berk *et al.*, 2001). Children, adolescents and adults have reported that their self-confidence has been affected by having a CLP (Noar, 1991; Turner *et al.*, 1997).

Body image and satisfaction with facial appearance.

Although children and adults with CLP seem relatively satisfied with their body image (Clifford *et al.*, 1972; Brantley and Clifford, 1979a; Marcusson *et al.*, 2002), some features specifically associated with CLP, such as the nose and teeth, are considered less than satisfactory (Tyl *et al.*, 1990; Marcusson *et al.*, 2002), and may result in problems with body image (Bernstein and Kapp, 1981).

While a number of studies suggest that those with CLP are generally pleased with their facial appearance (Clifford *et al.*, 1972; Bjornsson and Agustsdottir, 1987; Slifer *et al.*, 2003), many express dissatisfaction (Richman, 1976; Kapp, 1979; Thomas *et al.*, 1997; Marcusson, 2001; Marcusson *et al.*, 2002), and desire further treatment to improve their appearance (Marcusson *et al.*, 2002). A visible scar appears to be of most concern, even to those with speech and hearing difficulties (Bernstein and Kapp, 1981). Little agreement has been reported between parents and children regarding satisfaction with facial appearance (Thomas *et al.*, 1997; Turner *et al.*, 1997; Slifer *et al.*, 2003).

Satisfaction with facial appearance has been linked to the incidence of behavioural problems (Clifford, 1969; Richman, 1983; Richman *et al.*, 1985).

Satisfaction with speech.

Few adolescents and young adults with CLP are dissatisfied with their speech (Noar, 1991). However, those with concerns regarding facial appearance are also more likely to have concerns regarding speech (Ramstad *et al.*, 1995b). Children with CLP have been shown to have poorer conversational skills compared with controls (Chapman *et al.*, 1998).

Associations have been made between behavioural problems and speech ability among children with CLP. Children with clefts who have normal speech tend to have fewer behavioural problems, as reported by their mothers (McWilliams and Musgrave, 1972). However, a longitudinal study found that at 9 years of age, children with fewer speech problems showed higher levels of internalizing behaviour (Richman, 1997).

Behavioural problems.

There are contrasting reports regarding behavioural problems among children with CLP. Behavioural problems have been reported among children with CLP, such as a tendency to have higher than average levels of internalizing behaviour, a risk factor for developing anxiety disorders (Harper and Richman, 1978; Richman and Millard, 1997). However, some studies have reported that children with CLP are less aggressive than controls (Starr, 1978) and do not exhibit any signs of delinquency (Bernstein and Kapp, 1981).

Parents of children with clefts have described their children as having more externalizing behavioural problems compared with parents of children without controls (Slifer *et al.*, 2003). Interestingly, one study found that parents of children with CLP were significantly more tolerant of behavioural problems in their children when compared with normative data (Tobiasen and Hiebert, 1984).

Social functioning.

According to professionals involved in caring for patients with clefts, many are socially affected as a result of having a cleft (Noar, 1992). Self-reports of adults with CLP also indicate a number of social problems (McWilliams and Paradise, 1973; Peter and Chinsky, 1974a). Differences have been established between young people with CLP and controls, such as those with CLP dropping out of school more frequently and being less likely to belong to clubs and societies (McWilliams and Paradise, 1973; Peter and Chinsky, 1975). A larger proportion of young people with CLP indicate no aspiration for further education when compared with controls (Peter and Chinsky, 1974b), and they are more frequently unemployed with a significantly lower income aspiration than controls (Peter *et al.*, 1975).

Two areas of social functioning appear to differentiate those with CLP from those without CLP: marriage and friendships. Fewer adults with CLP marry (McWilliams and Paradise, 1973; Broder *et al.*, 1994), and even when they do, marriage occurs later in life (Peter and Chinsky, 1974a; Broder *et al.*, 1994). Childless marriages occur more frequently in subjects with clefts than among controls (Peter and Chinsky, 1974a). Studies have suggested that children and young adults with CLP have fewer friends than non-cleft controls (Noar, 1991; Ramstad *et al.*, 1995a; Bressman *et al.*, 1999), with only one study suggesting that children with clefts have more friends than control children (Broder *et al.*, 1994). However, the authors of that study urged caution when interpreting the unusual finding.

Anxiety and depression.

Anxiety and depression have been reported to be twice as prevalent in adults with CLP compared with normal controls (Ramstad *et al.*, 1995b). Dissatisfaction with facial

appearance has been found to be a predictor of depression among subjects with clefts and controls (Marcusson *et al.*, 2002). Berk *et al.* (2001) examined social anxiety among Chinese adults with CLP and found significantly more social anxiety and avoidance among those with a CLP than among siblings and controls.

Attachment, development and learning.

Few differences exist between children with CLP and controls regarding mother–child attachments (Hoeksma *et al.*, 1996; Speltz *et al.*, 1997; Maris *et al.*, 2000) and interactions (Endriga and Speltz, 1997). Maris *et al.* (2000) reported lower attachment security at 12 months of age. However, this was not the case by 24 months of age.

Less attractive infants (regardless of whether or not they have a cleft) are more likely to have secure attachments (Speltz *et al.*, 1997). This finding is acknowledged as unusual by Speltz *et al.* (1997) in the light of previous research purporting to agree with the ‘what is beautiful is good’ hypothesis. In an attempt to understand the unusual finding, Coy *et al.* (2002) replicated the study and came to similar conclusions. The authors of both studies suggest that it may be appropriate to adopt an ‘infant vulnerability’ hypothesis when interpreting the findings. This suggests that the infant’s secure attachment is accounted for by the mother’s increased protectiveness of her child with less attractive facial characteristics (Speltz *et al.*, 1997; Coy *et al.*, 2002).

Differences have been established regarding scores on mental development with babies as young as 5 months old with clefts compared with normative data. At 5 months of age, babies with clefts were found to be at risk of delayed development (Neiman and Savage, 1997). However, at 36 months this difference appears to dissipate, except in one area, expressive language. Starr *et al.* (1977) examined the mental development of infants using cross-sectional and longitudinal data. The cross-sectional data showed some differences between cleft groups and normative data regarding mental development, but this was not the case when the data were analysed longitudinally. Kapp-Simon and Krueckeberg (2000) carried out a similar study and found that mental development scores decreased significantly as infants with CLP increased in age.

A number of studies have reported specific learning problems among children with CLP (Brantley and Clifford, 1979a; Millard and Richman, 2001), with one study finding that one in four children with a cleft repeated a grade at school (Broder *et al.*, 1998).

Is there a relationship between cleft type and the prevalence and severity of psychosocial impairment?

The type of cleft and its severity appears to have little influence on the individual’s overall psychosocial functioning. No significant associations have been found

between cleft type and the incidence of behavioural problems (Richman, 1976, 1978; Starr, 1978, 1980a; Speltz *et al.*, 1993; Richman and Millard, 1997; Millard and Richman, 2001), self-esteem (Starr, 1978, 1980a, b), mental and motor development (Starr *et al.*, 1977; Tyl *et al.*, 1990), or psychosocial functioning in general (Heller *et al.*, 1981). However, a few differences between cleft types have been found in relation to self-concept, satisfaction with facial appearance, depression, attachment, learning problems and interpersonal relationships.

CLP.

Compared with control children, children with CLP have been shown to have the lowest self-concept scores of any type of cleft (Broder and Strauss, 1989). Those with visible defects (i.e. cleft lip or CLP) have expressed greater dissatisfaction with their appearance than those without a cleft lip (Broder *et al.*, 1994; Thomas *et al.*, 1997). Subjects with CLP were found to marry later than those with any other type of cleft (Peter and Chinsky, 1974a). However, levels of depression were lower among children with unilateral CLP compared with those with cleft palate only (CPO) or bilateral CLP (Millard and Richman, 2001).

CPO.

In those with CPO, infants display lower rates of attachment security than infants with CLP or controls at 12 months of age (Maris *et al.*, 2000). However, by 24 months this difference seems to dissipate. Endriga and Speltz (1997) noted that mothers of babies with CPO appeared less involved in mother–infant face-to-face interaction than mothers of babies with CLP.

Learning problems appear to be more prevalent among children with CPO in comparison with other cleft types (Broder *et al.*, 1994, 1998; Millard and Richman, 2001), especially in males (Broder *et al.*, 1998). Children with CPO had greater problems with parent and teacher reported anxiety and depression compared with children with either unilateral or bilateral CLP (Millard and Richman, 2001). On a more positive note, males with CPO tended to follow a similar pattern to siblings and controls in terms of marriage status, age at first marriage, and the number of children within a marriage (Peter and Chinsky, 1974a).

Are particular age groups more vulnerable to psychosocial problems?

The age of an individual with CLP does not appear to influence the occurrence or severity of psychosocial problems. However, there are a few exceptions to this. Increasing conduct problems have been reported as the child with CLP grows older (Schneiderman and Auer, 1984). In relation to speech, parents and children with CLP have expressed increased satisfaction as the child gets older

(Broder *et al.*, 1992). While individuals of all ages with CLP have reported dissatisfaction with their facial appearance (Kapp, 1979; Ramstad *et al.*, 1995b; Thomas *et al.*, 1997; Marcusson *et al.*, 2002), one study did find that younger children and adolescents (aged 10 and 15 years) were more dissatisfied with their appearance than subjects aged 20 years (Thomas *et al.*, 1997). Satisfaction with appearance in females has been found to decrease with age among those with inter-oral defects only, i.e. isolated cleft palate (Broder *et al.*, 1994).

Discussion

A systematic review normally involves the use of a meta-analysis of the studies included. However, it was clear that the large variation in study design and outcome measures adopted by previous researchers meant that a collective examination using the meta-analysis technique was not possible. The results and conclusions of this review are therefore presented by focusing on any statistically significant findings reported by the authors in relation to psychosocial problems among those with CLP as they appeared in the original papers.

This review has found that conflicting evidence exists in the literature when it comes to establishing whether children and adults with repaired CLP experience increased psychosocial problems as a result of their cleft. Some areas of psychosocial functioning (for example, behavioural problems or satisfaction with facial appearance) appear to pose difficulties for those with CLP. However, the nature of the studies that have been conducted in this area made it impossible to apply statistical techniques to assimilate the data. On the basis of currently available evidence, it is impossible to state the extent of the problem with any certainty. For every study reporting psychosocial problems among those with CLP, there are others which refute this finding. Where difficulties have been established, they are mostly related to behavioural problems, dissatisfaction with facial appearance, and difficulty with specific aspects of social functioning. Self-esteem among subjects with CLP is considered to be generally good and in some cases is unusually high. However, unusually high scores can, in certain circumstances, be indicative of self-esteem problems. Very high scores can indicate a socially desirable response pattern, skewed self-perceptions, or a deliberate attempt to present a very positive image (Brown and Alexander, 1991), and should be investigated further.

Associations between cleft type and the increased incidence of certain psychosocial problems have been established in relation to CLP (e.g. dissatisfaction with appearance and marrying later in life) and CPO (e.g. insecure attachments and learning problems). However, the evidence is not strong, making it difficult to state definitively that one type of cleft results in more severe psychosocial problems than another type. Further research is needed in all areas of

psychosocial functioning, with data analysed according to cleft type.

Lack of appropriate controls

Studies that employ appropriate control groups are obviously preferable to uncontrolled studies and produce more convincing results. Unfortunately, more than half of the studies included in this review did not use a control group. In a number of investigations, normative data or data from previously conducted national studies were employed (Starr *et al.*, 1977; Tobiasen and Hiebert, 1984; Geier and Wittstock, 1986; Tyl *et al.*, 1990; King *et al.*, 1993; Ramstad *et al.*, 1995a, b; Bressman *et al.*, 1999; Cochrane and Slade, 1999). Contemporaneous controls are essential so that children with CLP are compared directly with their peer group, thereby reflecting the subtle societal and cultural influences that often affect how an individual's facial appearance is perceived.

Lack of longitudinal studies

The results of the investigations included in the review do not help to identify the severity and duration of psychosocial problems among those with CLP. Only nine studies in this review presented longitudinal data (Starr *et al.*, 1977; Heller *et al.*, 1985; Hoeksma *et al.*, 1996; Jocelyn *et al.*, 1996; Richman, 1997; Richman and Millard, 1997; Speltz *et al.*, 1997; Maris *et al.*, 2000; Coy *et al.*, 2002). Unfortunately, the results of these nine studies do not help to clarify the extent of the psychosocial problems. The studies measured different psychosocial constructs ranging from attachment to behavioural problems and focused on different age ranges, making it difficult to draw conclusions.

Longitudinal studies involving subjects with clefts and age- and sex-matched controls are needed to identify the duration of psychosocial problems and to determine whether specific developmental stages are influential in the occurrence of these problems. The majority of studies in the review focused on children and adolescents with CLP. It is important to determine whether problems experienced by children with CLP continue into adulthood or perhaps only emerge in adulthood.

Longitudinal studies could take into account the influence of facial growth, which is not complete until the late teenage years, and children and adolescents with CLP may undergo a number of surgical procedures during this time. Facial growth changes and the experience of surgery should be examined as possible factors influencing psychosocial functioning. In particular, facial appearance may change as a result of growth or treatment interventions and, depending on whether these changes are pleasing or otherwise to the individual, improvements or deterioration in psychosocial functioning may result. If we are to identify those most at risk from psychosocial problems

and to determine the extent of such problems, these factors require more investigation.

Research methods

The studies included in this review employed a wide variety of questionnaires ($n = 67$), making it difficult to directly compare the results. In addition, many of the questionnaires used were originally designed up to 30 years ago, which may reduce the validity of the comparative normative data in the contemporary setting.

While there appears to be ample research regarding psychosocial functioning among those with CLP, only a few studies draw conclusions concerning each major area of functioning. It would appear that attachment among infants with clefts and their mothers, and the mental development of infants with clefts, are the only areas where studies have been replicated. Replicating previous studies would assist in drawing conclusions regarding the occurrence of psychosocial problems.

Even among studies measuring similar aspects of functioning, the results are contradictory. This lack of conclusive evidence is hardly surprising given the absence of uniformity in the methods used. Multicentre research using uniform and agreed methodology is needed to enable a more useful body of evidence to emerge.

The reviewed studies employed a mixture of self-report, parent, teacher or professional reports. However, 21 of the studies did not include the self-report of the individual with CLP. The results of these studies should be considered in the light of other findings that show poor agreement between parent and patient pairs (Thomas *et al.*, 1997; Turner *et al.*, 1997).

Of the 64 studies included in the review, 31 were published before 1990. Surgical techniques in the area of CLP have become more refined in the past 30 years and therefore the visible deformity associated with this anomaly may not be as pronounced today. In addition, there is a greater awareness of the difficulties associated with being visibly different and it is likely that the clinician's approach to such children and adults has changed for the better.

The ability of some children with CLP to do better than others may be explained by the concept of resilience. Strauss (2001) suggested that the theories of individual and family resilience are important perspectives when investigating the lives of those with CLP. Most of the studies in this review lacked measurement of variables that may explain why some children with CLP do better than others, such as family support and the coping skills of the child. These factors may account for a lack of evidence in the literature indicating serious psychosocial problems as a result of CLP. However, this needs to be investigated in light of the recent improvements in cleft care. Positive aspects to facial difference (Eiserman, 2001), awareness of the damage of stigmatizing children with craniofacial conditions (Kelton,

2001), and a shift in attitude towards the strengths rather than the deficits of the child (Mouradian, 2001) are suggested as the way forward in conducting research in this population.

The vast majority of studies did not include an interview with the affected individual. In the absence of specific psychosocial measures for children and adults with CLP, more studies could include a semi-structured interview to ensure topics relevant to CLP are included. Of particular interest should be the individual's coping skills, which may reveal why some individuals encounter psychosocial problems as a result of CLP and others do not. These issues could be more fully addressed and understood during the interview process. Findings from interview data could then be used to design measures specifically for use with this patient group.

Conclusion

Despite finding a relatively large volume of literature in this area, the studies lack uniformity and consistency to adequately evaluate the psychosocial problems resulting from CLP. Although there is some limited evidence to suggest that individuals may encounter psychosocial problems as a result of having a CLP, overall adjustment and functioning appear to be reasonably good. However, this cannot be stated with any certainty until decisions are based on a higher quality evidence base than is presently available.

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Appendix

Keywords used in the literature search

Cleft, lip, palate, CLP, palatal, psychology, psychological, psychosocial, psych\$, social, impact, influence, effect, problem, unilateral, bilateral, complete, primary, secondary, facial, orofacial, personality, adjustment, self, concept, esteem, image, body-image, appearance, confidence, speech, behaviour (*UK spelling*), behavior (*US spelling*), anxiety, mood, depression, achieve\$, attachment, develop\$, learning. \$ denotes unlimited truncation which retrieves all possible suffix variations of a root word.

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