

Critical Review: What are the Psychosocial Effects of Cleft Lip and Palate?

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This critical review examines the specific psychosocial effects that the diagnosis and treatment of cleft lip and palate has on individuals of all ages. Study designs include a systematic review, a case-control study and a retrospective chart review. Overall, the literature reviewed provides variable evidence regarding the topic of interest. It is thought that the diagnosis and treatment may have a negative impact on some individuals, although the extent and severity remain unknown. Recommendations for future research and clinical practice are provided.

Introduction

Cleft lip and palate (CLP) is a congenital malformation of the face and oral cavity that occurs during pregnancy (Kummer, 2008). Management of CLP is a lifelong process and requires multiple surgical and other intervention procedures to deal with issues such as facial abnormalities, difficulty feeding, speech impairments, abnormal resonance and hearing difficulties (Baker, Owens, Stern, & Willmot, 2008; Kapp-Simon, 2004; Kummer, 2008). In light of the complex nature of CLP, it is important to understand not only the medical implications of the disease and treatment, but the psychosocial implications as well (Cheung, Pheng, & Ho, 2006).

In recent decades, advances in the surgical technique and sequencing of procedures has allowed for improved repair of CLP. Similar advances have been made in the awareness, and attention to the psychosocial effects that CLP may have on individuals. With this has been a shift from focusing purely on the surgical treatment of CLP to a more holistic approach including geneticists, pediatricians, psychologists and social workers (Gussy & Kilpatrick, 2006; Kummer, 2008).

Research in the area of psychosocial effects of CLP leaves many questions unanswered. While the majority of studies report that CLP has a negligible psychosocial impact on an individual's overall well-being, specific psychosocial issues such as behavioural difficulties, dissatisfaction of facial appearance, withdrawal from social situations and symptoms of anxiety and depression have been identified (Brand et al., 2008; Gussy & Kilpatrick, 2006; Hunt, Burden, Hepper, & Johnston, 2005a). Several factors may have contributed to these mixed findings including differing definitions of the term "psychosocial", focusing on a single psychosocial construct which is then used to generalize about the individual's overall psychosocial well-being, lack of

longitudinal studies and control groups, and failure to distinguish between variations of cleft lip and or palate (CL/P) (i.e. cleft lip only (CLO), cleft palate only (CPO), cleft lip and palate (CLP) (Gussy & Kilpatrick, 2006; Hunt, Burden, Hepper, Stevenson, & Johnston, 2005b). Due to these methodological weaknesses, the specific psychosocial effects of CLP remain undetermined (Gussy & Kilpatrick, 2006).

For the purposes of this critical review, the term 'psychosocial' refers to an individual's psychological development and interaction with a social environment. Based on the articles included in this critical review, the following constructs are included: self-concept, body image, satisfaction with facial appearance, satisfaction with speech, behavioural problems, social functioning, anxiety, depression, attachment, development and learning (Brand et al., 2008; Hunt et al., 2005a; Snyder & Pope, 2009).

As members of the primary care team treating individuals with CLP, it is crucial that speech-language pathologists (SLP) remain informed and conscientious about all aspects of care, including the psychosocial implications of the disease and treatment. Although specific implications have yet to be determined, the evidence surrounding the psychosocial adjustment of individuals with CLP should be critically examined and evaluated to allow the discerning SLP to function effectively as part of the CLP care team.

Objectives

The primary objective of this paper is to provide a critical evaluation of the existing literature on the psychosocial effects of CLP of individuals of all ages. A secondary objective is to provide recommendations for clinical practice and areas for future research.

Methods

Search Strategy

Computerized databases, including CINAHL, SCOPUS, PubMed, and ProQuest were searched using the following search strategy:

(psychosocial) AND (cleft lip) AND (cleft palate).

The search was limited to articles published in 2005 or later as a systematic review provided critical analysis of articles published prior to 2005. In addition, articles were limited to those written or translated into English.

Selection Criteria

Studies selected for inclusion in this critical review paper were required to investigate the psychosocial effects on individuals with CLP. Cleft lip and palate, as opposed to CLO, CPO or CL/P, was chosen in an effort to highlight the visible and invisible effects of both cleft lip and cleft palate, as well as focus on a specific CL/P diagnosis (Joachim & Acorn, 2000).

Data Collection

Results of the literature search yielded the following types of articles congruent with the aforementioned selection criteria: systematic review (1), case-control study (1), and retrospective chart review (1).

Results and Discussion

Levels of evidence discussed below are based on the Oxford Center of Medicine, 2011.

Systematic Review

Hunt et al. (2005a) conducted a systematic review to determine whether the presence of CLP places an individual at an increased risk of having psychosocial problems. Results of the study revealed that although the majority of individuals with CLP do not experience major psychosocial problems, specific problems may arise including behavioural problems, dissatisfaction with facial appearance, depression and anxiety.

Clear guidelines of the authors' search strategy and selection criteria of articles was reported in detail. A total of 64 articles met the full inclusion criteria focusing on studies with individuals with repaired, non-syndromal CLP. All variations of the term CLP were searched, including cleft lip only (CLO), cleft palate only (CPO) and cleft lip and palate (CLP), with results reported separately for each cleft type. Due to the focus of this critical review, only the results and analysis of CLP will be reported. Individual studies were rated independently by two researchers to determine their suitability, with

96% agreement between the two reviewers. In addition, proformas were completed independently by two authors to identify specific features of each study (i.e. participants, age range, sample size, method of measurement, etc.), with complete agreement existing between the two authors. It is important to note that although researchers included a well-defined definition of psychosocial functioning, several studies included in the review examined only a single psychosocial or social construct, rather than psychosocial functioning in its entirety, limiting the interpretation of the conclusions with regard to psychosocial effects.

Although Hunt et al. (2005a) clearly described their search strategy and selection criteria, the researchers' method of analysis was not described clearly or in sufficient detail such that the methods could be well understood and the study could easily be replicated.

Researchers acknowledged several limitations within their study. First, more than half of the articles included in the review did not use a control group, leaving subtle societal and cultural influences on an individual's psychosocial wellbeing unrevealed. Secondly, there were not sufficient similarities in the populations studied or the measurements used; therefore a traditional systematic review was not possible. In addition, 31 of the 64 articles included in the review were published before 1990. With advancing technology and refinement of surgical techniques, the visible deformity associated with CLP may not be as significant today as it once was, limiting the current applicability of the results. Finally, effect size and confidence intervals were not reported for the data, limiting the interpretation of the systematic review's results.

Despite the numerous limitations within this systematic review, the article by Hunt et al. (2005a) is valuable as it provides suggestions for further research, such as longitudinal studies, adequate control groups and analyzing all data according to cleft type.

Given the level 1 design of the study and the analyses carried out, as well as the strengths and weaknesses present, these results can be interpreted with a suggestive degree of confidence. Therefore, it is recommended that these findings be considered during the diagnosis and treatment of CLP.

Study #1

Brand et al. (2008) conducted a case-control study to investigate the psychosocial functioning in children and adolescents with CLP. A case group of 32 CLP subjects was compared to an age- and gender-matched control group of 34 non-CLP subjects. Results of the study indicated that subjects

with CLP were six times more likely to report difficulties participating within the general social environment and in proactive behaviour, although no differences were noted during interaction with family, friends or peers, or with respect to emotional problems, conduct problems or hyperactivity.

In addition to focusing on psychosocial functioning, the article's main purpose was to examine how CLP and the psychosocial effects that result affect sleep and sleep patterns. In accordance with this paper's focus, only the psychosocial effects of CLP will be reported.

Information on subject selection was limited. While Brand et al. (2008) reported that the target group (CLP group) and control group were age- and gender-matched, there was no data on subjects' age, type of cleft (i.e. unilateral, bilateral), severity of cleft or history of psychosocial disturbances, all important control variables. In addition, all CLP subjects were recruited from the same clinic and underwent the same surgical CLP repair. Although this allowed researchers to maintain consistency of participant experience, it also may have introduced bias and limited the study's ability to generalize results.

Brand et al. (2008) clearly defined the psychosocial areas investigated within each questionnaire, and provided adequate explanation of the proceedings such that the measures were understood and the study could be replicated. Both questionnaires, the Strength and Difficulties Questionnaire (SDQ), which looked at emotional symptoms, conduct problems, hyperactivity, peer problems and prosocial behaviour, as well as the Participation in Everyday Life Communication Questionnaire (PIELCQ), which looked at social environment, family, friends/peers and proactive behaviour, reported good reliability and internal consistency. It was acknowledged by researchers that the 3-point response scale format of the SDQ reduced the ability for more detailed data analysis. Similarly, researchers acknowledge that the PIELCQ is currently in a stage of evaluation and optimization with validity not yet established; therefore results should be interpreted with caution.

Appropriate statistical analyses were conducted: t tests and Chi-squares were used to analyze demographic differences between the target and the control group. Analysis of Variance (ANOVA) was conducted to evaluate the effects of age or CLP status (participants with or without CLP) for dimensions on the SDQ and PIELCQ. In addition, a confirmatory factor analysis with orthogonal rotation was conducted to determine psychosocial functioning variables, as an initial step for multiple linear regression to calculate the influence on sleep. It

should be noted that confidence intervals were not reported for data on psychosocial functioning and CLP, therefore the reader's interpretation of the statistical results is limited.

Given the level 2 design of the study and the analyses carried out, as well as the strengths and weaknesses present, these results can be interpreted with a suggestive degree of confidence. Therefore, it is recommended that these findings be considered when diagnosing and treating individuals with CLP.

Study #2

Snyder and Pope (2009) conducted a retrospective chart review to examine the psychosocial effects of craniofacial anomalies (CLP, CLO, CPO, nonsyndromal craniosynostosis, hemifacial microsomia and hemangioma) on children and adolescents. A total of 408 patients included in the study, with 174 patients aged 2-18 with a CLP diagnosis, were compared to published norms of non-referred, healthy children. In light of the focus of this critical review, results will be reported for children with CLP only, despite the additional craniofacial anomalies included within the study. Results of the study indicated that children with CLP showed elevations into the clinical range with regards to social problems, school capability and activities when compared to other craniofacial diagnosis groups and these difficulties increased with age, although there was little evidence of true psychological disorder in any age group.

The selection criteria for CLP participants was reported in detail, with data on participants' age, gender, diagnosis and informant (i.e. mother, father, other) who completed the protocol. A clear description of the published norms was not included, with only a reference given to access further information. The author obtained the article that was referenced and determined that a representative sample was used. It is important to note that all CLP participants were patients of the same reconstructive plastic surgery department at an urban medial center, which may have introduced bias and limited the study's ability to generalize results to other populations.

Snyder and Pope (2009) defined the psychosocial areas investigated within the questionnaire, and provided adequate explanation of the proceedings such that the measures were understood and the study could be replicated. The Child Behaviour Checklist-2/3 (CBCL-2/3) was reported to have adequate validity and reliability, while the Child Behaviour Checklist- 4/18 (CBCL-4/18) was reported to have been used extensively with normative and clinical samples and had excellent validity and reliability. Although a minor

critique, researchers did not define the descriptors 'adequate' and 'excellent' thus leaving readers with a vague understanding of these terms. It was acknowledged by researchers that while the use of parent-completed protocols for all ages provided a useful perspective, self-reports would allow for a more complete understanding of psychosocial functioning of this population. As a result, this study is not able to distinguish true psychosocial impairments from parental perceptions and inferencing.

Independent t-test analyses were conducted separately based on diagnosis and age group, allowing readers and other researchers to obtain detailed data on each anomaly studied, rather than as an entire group. Following, chi-square analyses were conducted to determine the presence of significant differences between the CLP group and normative samples. Researchers reported that boys and girls were combined within diagnoses to increase the sample sizes of each group, however, a better rationale for combining groups would be based on the assumption that boys and girls do not differ in the psychosocial effects of CLP. Finally, effect size and confidence intervals were not reported for the data, limiting the interpretability of results by the reader.

Given the level 2 design of the study and the analyses carried out, as well as the strengths and weaknesses present, these results can be interpreted with a suggestive degree of confidence. Therefore, it is recommended that these findings be considered when diagnosing and treating individuals with CLP.

Conclusion and Clinical Implications

Due to the moderate strength of the evidence presented in the three articles reviewed, it is recommended that clinicians remain cautious when implementing the findings of these studies into clinical practice.

While the current critical review does not provide strong evidence regarding the relationship between negative psychosocial effects and the presence of CLP, clinicians should remain mindful of the heterogeneity of the CLP population as well as extraneous variables that may influence an individual's psychosocial well-being. Therefore, the diagnosis and treatment of CLP may negatively impact some individuals, although the extent and severity remain unknown.

Recommendations

It is recommended that further research be conducted to clarify and confirm the specific psychosocial effects that CLP have on an individual. In order to

improve the level of evidence provided by the existing literature, it is recommended that the following are taken into consideration:

- a) Future research should employ longitudinal study designs in order to capture a full view of the psychosocial effects across the lifespan at each stage of development, allowing the results to be more applicable to clinical practice
- b) Future studies should provide a clear and concise definition of 'psychosocial' to allow for consistency and comparison with additional literature.
- c) Future research should employ a qualitative methodological approach (e.g. narrative) to elicit some lived experiences of individuals with CLP. This approach would allow for a richer, deeper understanding of potential impacts of CLP psychosocially. Though not generalizable, these rich understandings can afford clinicians new insights and more sensitive practices.
- d) Researchers should distinguish between variations of CL/P in order to identify psychosocial implications specific to the each CL/P diagnosis.
- e) Including appropriate control groups would be beneficial in further research to allow for more detailed comparison and analysis among groups.
- f) SLPs, along with other professionals involved in CLP care, must acknowledge and remain cautious of the possible psychosocial implications of CLP in order to preemptively identify and treat if these problems may arise.

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