Critical Review:

Do the reading skills of children with nonsyndromic cleft lip with or without cleft palate resemble their noncleft peers?*

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This critical review examines the literature on the reading skills of children with and without nonsyndromic cleft lip and palate. All four articles included in this review had a between groups study design. Overall, the results of this review provide suggestive evidence that children with nonsyndromic cleft lip with or without cleft palate score lower on measures of reading as an overall group however the evidence does not suggest that this group is scoring within the deficit range. Recommendations for clinical practice and future research are discussed.

Introduction

Orofacial clefts occur as a result of a disruption in the embryological development of the structures of the face (Bender, 2000). According to Bender (2000), cleft lip with or without cleft palate (CL/P) is one of the most common structural birth defects occurring in approximately one in seven hundred newborns. The presentation and severity of the cleft varies considerably based on the location of the structural deficit and the extent of tissue involvement (Bender, 2000). For example, the cleft may involve the lip only, the palate only, or a combination of both. Additionally, clefts can occur unilaterally, affecting only one side of the face, or less commonly bilaterally, affecting both sides. Clefts can also be classified as incomplete or complete based on the number of structures affects (Bender, 2000). A distinction is also made between syndromic and nonsyndromic cleft lip with or without cleft palate (NCL/P) which is of significance for the present review. Bender (2000) defines NCL/P as the absence of any physical or developmental anomalies except for the orofacial cleft. The cause of NCL/P is considered to be multifactorial with possible genetic and environmental predecessors (ASHA, 2016).

Diagnosis of a cleft lip is possible prenatally through ultrasound imaging as early as 13 weeks (ASHA, 2016). Prenatal detection of cleft palate however is difficult due to visual restrictions (Bender, 2000). Identification of cleft palate is usually made after birth and depending on the severity can be missed until feeding difficulties are expressed (Bender, 2000). Treatment protocol for individuals with CL/P requires them to be followed by a multidisciplinary craniofacial team. Treatment involves multiple orofacial surgeries including reconstructive and dental surgeries (Bender, 2000). Compensatory errors or resonance issues may become apparent due to structural insufficiencies and speech therapy or additional surgery may be recommended (ASHA, 2016). Additionally, children with cleft palate commonly experience recurrent middle ear infections causing a temporary reduction in hearing ability which can impact the child's ability to learn speech and language (ASHA, 2016).

A relationship between speech, language, and reading has been reported and is well documented in the literature (Chapman, 2011). Research has reported the existence of both speech and language delays in children with CL/P (Chapman, 2011). The current view of reading is that it is a "language-based" skill (Chapman, 2011). Several explanations have been offered as to why children with nonsyndromic orofacial clefts may have an elevated risk for reading difficulties. Research has suggested possible neurological differences between children with and without orofacial clefts proposing a possible biological vulnerability (Collett, Stott-Miller, Kapp-Simon, Cunningham, and Speltz, 2010b). For example, functional imaging research has reported that children with orofacial clefts exhibited neural inefficiency during reading and language tasks (Collett et al., 2010b). Others have hypothesized that perhaps it is the cleft itself that places the child at risk because of early speech deficits as normal anatomy and physiology are considered prerequisites for language development (Collett et al., 2010b). Others still have theorized that the presence of frequent middle ear infections may offer an explanation as hearing deficits can impact language acquisition and specifically phonological development (Collett et al., 2010b). Children in kindergarten through second grade are of particular interest because research has found this time period to be a critical period for identifying and intervening with reading problems (Collett et al., 2010b).

Objectives

The primary objective of this review is to critically evaluate existing literature regarding the impact of nonsyndromic cleft lip and/or palate on reading skills in children. The secondary objective of this paper is to provide evidence-based recommendations for clinical practice and future research.

Methods

Search Strategy

Journal articles related to the topic of interest were located using the following computerized databases: SCOPUS, PubMed, and Google Scholar. Databases were searched using the following key terminology: [((orofacial cleft) OR (cleft) OR (cleft lip and palate)) AND ((reading) OR (reading disability) OR (literacy)) AND ((child*) OR (school age*))]. Reference lists of previously searched articles were used to obtain additional related studies.

Selection Criteria

Studies selected for inclusion in this critical review were required to investigate the impact of nonsnydromic cleft lip and/or palate on reading skills in children between the ages of 5-7 years. No limits were set on the study design, demographics, or date of publication. Studies including syndromic cleft lip and/or palate were excluded.

Data Collection

The results of this literature search yielded four articles congruent with the selection criteria. All selected articles had a between groups study design which was determined to be an appropriate design for the question being addressed.

Results

Richman, Eliason, and Lindgren (1988) conducted a between groups study to examine reading disability in elementary school students with cleft lip and palate (CLP) and cleft palate only (CPO). Recruitment and exclusionary criteria were described in detail. Reading disability was defined as obtaining a standard score below one standard deviation of the mean. A group of 172 children between the ages of 6-13 years were separated into groups based on having CLP or CPO and age. Although both CLP and CPO were of interest to the present study, only the youngest groups were within the age range of interest to the present review (6-7 years). For the purposes of this paper, the distinction between cleft lip and palate (CLP) or cleft palate only (CPO) was not made. Inclusion criteria ruled out children with mental retardation, as well as those with behavioral and emotional difficulties. Reading was evaluated using a published reading test with clinical rather than norm based data that measured word recognition and reading comprehension. Richman et al. (1988) provided adequate information regarding procedures such that the measures were well understood and provided sufficient detail for replication. Results revealed a high rate of reading disability overall and a somewhat higher rate for the youngest group. Of the children in the youngest age category, 48-53% were identified as having a reading disability.

Several limitations existed within this study. A major limitation was the lack of a typically developing control group which made interpretation of results difficult. In addition, the use of a norm referenced test rather than a criterion referenced test would have also improved the interpretability of the results. Another limitation was the sample population from which the data was collected. This population was made almost entirely of Caucasian monolingual speakers making generalization of the results to the broader population difficult. As well, some of the sample received speech or learning disability services which could have skewed the results by reducing the percentage of children presenting with a reading disability.

Overall, the study provides somewhat suggestive evidence of reading disability among children with cleft. This study was limited by its lack of a control group and norm referenced test. The findings regarding higher rates of reading disability in the youngest group must be interpreted with caution.

Collett, Leroux, and Speltz (2010a) conducted a longitudinal case control study to compare the reading and language skills of children with nonsyndromic orofacial clefts (n=57) to those of a matched control group (n=77) from infancy through to age 7 years. For the purposes of this review, only the findings of reading achievement collected at age 7 years will be discussed. The selection criteria for both case and control subjects was reported in detail, with case controls matched according to socioeconomic status (SES), gender, and age. Recruitment occurred before the possibility of known or suspected reading problems was determined, reducing the potential for sampling bias. Children were evaluated using a published reading test that measured letter knowledge, single word reading, reading comprehension, and spelling ability, all of which were found to be highly correlated. Tests were administered by trained examiners, in accordance with standardized instructions. Interactions were videotaped and coded by trained observers and acceptable reliability was reported. A composite standard score was the developed and compared to controls. This composite score was

developed to reduce the number of group comparisons thereby decreasing the probability of a type one error. Appropriate statistical tests were conducted (t-tests) based on the normalized data. Results revealed that although both groups scored within normal limits, children with orofacial clefts scored higher than unaffected controls.

This study had several limitations which should be noted. Children were recruited from a single site which is problematic as it decreases the generalizability of the results to the larger population. Additionally, as a result of this single site recruitment procedure, white middle class children were overrepresented. This subgroup may be at a lower risk for reading impairments compared to children from a lower socioeconomic status. Further, attrition rates at age 7 were high (30%), which limited the statistical power to detect differences at this age. However, by using inverse probability weighting the authors determined that group differences did not appear to be influenced by attrition. Information on cleft severity or the number of associated minor physical anomalies were not included. A fundamental limitation of this study was that the standardized tests used to assess early reading skills did not measure key early indicators of reading development such as phonological processing, phonological memory, and rapid naming. These early indicators may be more sensitive to detecting group differences in young children and as such excluding them may have skewed the accuracy of the results. Also of importance, approximately one third to one half of the children with orofacial clefts had received speech intervention which may have reduced disparities in basic reading.

This study provides suggestive evidence that the reading levels in children with NCL/P and typical developing children are not different. Findings regarding higher scores in the CLP group however must be interpreted with caution due to the limited number of reading measures employed.

Chapman (2011) conducted a mixed case control study to investigate the relationship between early reading, communication skills, and cleft lip and/or palate (CLP). As this study is comparing two different populations it precludes randomization and thus a nonrandomized control trial is appropriate. A case group of 28 children 5-6 years and matched controls were recruited through well-specified channels. The case control group was matched according to age, gender, and months of formal schooling. Chapman (2011) provided adequate information regarding the procedures such that the measures were well understood and described in sufficient detail for replication. Commonly used standardized tests of reading, articulation, and oral language were used, of which the reading measure is relevant to the current review and will be discussed here. Assessments were scored according to the instructions in the test manuals. Multiple independent-sample t tests comparing groups revealed a significantly lower reading score in the affected group. Additionally, children with CLP preformed significantly poorer on an Alphabet subtest suggesting that they were behind their noncleft peers in letter-sound knowledge. Although children with CLP exhibited poorer performances on reading and reading related skills, results indicated that their mean standard score fell within the normal range.

Limitations of this study relate to aspects of methodology with which the study was conducted. Each participant completed a battery of standardized assessments that lasted approximately 2.5-3 hours. The authors did not specify order of completion. As a result, assessments completed at the end of the assessment period may not be representative of the child's abilities. Administering the assessments in random order or counterbalancing the order where half of the sample completed the assessments in one order and the other half in another order would aid in eliminating bias due to order effect. Additionally, the influence of multiple assessments on the probability of type 1 error was not accounted for.

Given the strengths and limited weaknesses of Chapman's (2011) study, the evidence presented is suggestive. The findings suggest that children with CL/P score lower on measures of reading as a group but fail to fall in the deficit range.

Collett, Stott-Miller, Kapp-Simon, Cunningham, and Speltz (2010b) conducted a between groups case control study to examine reading and related skills of children with nonsyndromic orofacial clefts (n=42) to those of a matched control group (n=43). Recruitment and selection criteria for both case and control subjects were reported in detail, with case controls matched according to age, demographics, sex, socioeconomic status (SES), months of school, and minutes of shared oral reading per week. Procedures and measures were described in detail allowing for replication. Reading was evaluated using a published test measuring basic reading, phonological awareness, phonological memory, reading fluency, reading comprehension, and rapid naming. Child testing sessions were completed in a 1.5–2 hour block in a clinic environment and were videotaped for later review. Assessments were completed by trained psychometrists in accordance with standardized procedures, with breaks taken as needed. Auditory stimuli were presented using a tape recorder which increases the consistency of delivery across participants and strengthens the results. A series of

appropriate linear regression analyses comparing children with clefts to controls was completed and multiple comparisons were controlled for to decrease the probability of Type 1 error. Results revealed that children with clefts scored significantly lower than controls on measures of basic reading, phonological memory, and reading fluency although the mean scores fell within the average range on all measures.

This was a well-designed case control study that appropriately met its objective. One limitation of this study is that children from lower SES, which has been known to correlate with reading, were underrepresented. Additionally, use of educational services by the CLP sample may have attenuated case control differences by improving early reading.

Overall, this study provides suggestive evidence that children with NCL/P score significantly lower on tests of reading compared to noncleft peers while still scoring within the average range.

Discussion

Language and reading skills among children with nonsyndromic cleft lip with or without cleft palate is an emerging area of interest in the literature. The objective of this paper was to critically evaluate the existing literature regarding the impact of NCL/P on reading skills in children. The literature search yielded four articles that met selection criteria. Each of the four articles had a between groups study design which allowed for more accurate comparisons to be made between the studies. Collectively, the four studies reviewed provided suggestive evidence that children with NCL/P score lower on measures of reading. As an overall group however, it appears that the reading skills of children with NCL/P do not fall in the deficit range.

Inconsistencies were present between the studies. These inconsistencies may be due to a multitude of factors. Notably, all the studies in this critical review had limited sample size limiting the power of the study to find differences and possibly detecting only gross effects. It may also be that the effect size of the group difference is too small making it difficult to find evidence of a statistical difference between the two groups. This could lead to inconsistent findings even in studies with somewhat larger sample sizes. Additionally, we may see studies observing deficits like we did in this review simply due to the variability of this population. These children differ in their presentation, surgical management, family support among others things, all of which could have impacted the results. Lastly, inconsistencies may be a result of the methodologies employed by the studies themselves. For example, the four studies differed in their assessment measures used to determine the presence of a reading impairment.

Although the current evidence suggests that children with NCL/P alone do not have a reading deficit, it might be that secondary conditions (e.g., hearing loss), in addition to the orofacial cleft, results in a reading score within the deficit range. Middle ear infections are nearly ubiquitous among children with CLP (Bender, 2000). Excluding children with cleft lip and palate who have hearing difficulties then may have reduced the generalizability of the results to the general population of children with clefts.

Conclusion

This critical review provided suggestive evidence that children with NCL/P score lower on measures of reading compared to matched peers while still maintain scores within the normal range.

Due to the inconsistencies found in the literature, this clinical question warrants further investigation. Future research considerations should include the following:

- Multi-site studies with larger sample sizes in order to increase generalization and extrapolation of results to the larger population;
- Appropriate and reliable assessment measures that examine key early indicators of reading development (e.g., phonological processing, phonological memory, and rapid naming) and are sensitive to detecting group differences in young children;
- Inclusion of secondary conditions such as frequent middle ear infections to determine if including these additional features results in children with NCL/P being identified within the deficit range for reading ability.

Clinical Implications

Children with orofacial clefts require several different types of services and as such, a multidisciplinary approach is required (ASHA, 2016). Early screening and intervention is important for identifying and treating all aspects of an orofacial cleft. These children are often followed by a craniofacial team for the first 18 years of life during which time ongoing management of all aspects of their cleft takes place.

This critical review provided suggestive evidence that children with NCL/P are scoring lower than their peers on tests of reading. Based on these findings and the

recommendations from ASHA (2016), every child with a orofacial cleft should be evaluated and monitored by a speech language pathologist on a consistent basis throughout their development. By doing so, intervention can be implemented at the earliest possible time. This is critical as research has indicated that intervention for reading difficulties ideally will start before 7 years of age (Collett et al., 2010b).

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