Critical Review:
Do the language skills of toddlers with nonsyndromic cleft lip and/or palate differ from their noncleft peers?

Sophia Ykema
M.Cl.Sc (SLP) Candidate
Western University: School of Communication Sciences and Disorders

This critical review examines the literature on the expressive and receptive language skills of toddlers with nonsyndromic cleft lip and/or palate. A literature search yielded one informational review and four mixed design studies. Overall, the results of the review provide highly suggestive evidence that toddlers with nonsyndromic cleft lip and/or palate score significantly lower on expressive language measures than age matched peers. The results for receptive language were mixed. Recommendations for clinical practice and future research are discussed.

Introduction

Clefts of the lip and/or palate are congenital malformations that occur in utero during the first trimester of pregnancy due to a disruption in embryological development. The developing structures of the lips, nose, hard palate and soft palate fail to properly fuse together at midline, resulting in open gaps of varying degrees in these anatomical facial regions. Clefts can be classified based on multiple factors and can occur in the lip only, the palate only or in both the lip and palate (Bender, 2000). Distinctions are made between unilateral clefts, affecting one side of the face, and bilateral clefts, affecting both sides of the face. Clefts are further described as either complete or incomplete, depending on the number of structures affected. Lastly, clefts can be divided into nonsyndromic and syndromic forms. Children with nonsyndromic clefts have no known teratogenic exposure that may cause cleft lip and palate and no other developmental or physical abnormalities (Murray, 2002). Individuals with cleft lip and/or palate are typically followed by a craniofacial team for the first eighteen years of their life. Remediation often requires multiple medical and non-medical interventions (Bender, 2000).

Children with cleft lip and/or palate may experience feeding issues, speech impairments, abnormal resonance, hearing difficulties and aesthetic problems. In addition to these, several explanations have been offered as to why children with nonsyndromic cleft lip and/or palate (NCL/P) may also have an elevated risk for early language delays. Although disagreement exists in the literature concerning the relationship between middle ear infections and language development, some authors have postulated that recurrent cases of middle ear infections may explain why some children with craniofacial clefts have delayed language skills (Casby, 2001; Zumach, Gerrits, Chenault, & Anteunis, 2010 Meinusch & Romonath, 2011). A second explanation is that the delayed development of the phonetic-phonological system in children with orofacial clefts interrupts the development of receptive and expressive language abilities (Hardin-Jones & Chapman, 2014). A connection between mothers with a direct style of interaction and delayed language skills in children with cleft lip and palate has also been reported in the literature (Meinusch & Romonath, 2011).

Early language delays can have detrimental effects on a child's ability to communicate with others and can lead to future behavioural, reading and writing difficulties (Jocelyn, Penko & Rode, 1996; Meinusch & Romonath, 2011). It is therefore paramount that Speech-Language Pathologists are aware of the language difficulties that may be associated with NCL/P so that they can best serve the needs of their clients.

Objectives

The primary objective of this review is to critically evaluate existing literature in order to determine whether the receptive and expressive language skills of toddlers with NCL/P differ from their noncleft peers.

Methods

Search Strategy

Online databases including PubMed, Google Scholar and Taylor & Francis were searched using the following keywords:

[((orofacial cleft) OR (cleft palate) OR (cleft lip and palate)) AND ((expressive language) OR (receptive language) OR (language development)) AND ((child) OR (toddler)).

Reference lists of relevant articles were used to obtain other related studies. The search was limited to articles available in English.

Selection Criteria

Papers selected for inclusion in this critical review were required to describe the early language skills of toddlers under the age of 30 months with NCL/P. Studies were excluded if they included participants with syndromic cleft lip and/or palate.

Studies that evaluated other factors (e.g. cognition, speech development) in toddlers with NCL/P were included, but for the purposes of the present review only information related to receptive and expressive language development will be discussed.

Data Collection

Results of the literature search yielded five research articles. One article was an informational review of the literature and four articles employed mixed designs.

Results

Informational Review of the Literature

An informational review of the literature provides an overview of the existing research on a specific topic. Most informational reviews do not provide information regarding how evidence is weighted and selected for inclusion. Therefore, these studies offer a lower level of evidence.

Hardin-Jones and Chapman (2011), two recognized experts in the field, published an informational review in order to summarize the evidence concerning the early language and cognitive abilities of children with nonsyndromic cleft palate. While there was no information listed concerning how the articles were selected, it appears that the studies included in the review represent a comprehensive list of the available evidence.

The authors noted that disagreement exists within the literature concerning whether toddlers with nonsyndromic cleft palate exhibit delays in receptive and expressive language skills. The majority of studies that focus on receptive language conclude that while toddlers with nonsyndromic cleft palate often perform worse than their noncleft peers, there is no significant difference between the groups. In terms of expressive language, the evidence suggests variable results with some toddlers with nonsyndromic cleft palate showing delays and others showing typical language development. Despite the limitations of informational reviews of the literature, this study offers suggestive evidence that the receptive language skills of toddlers with nonsyndromic cleft palate are often in the typical range whereas expressive language skills show more variability with some nonsybdrimnic cleft palate toddlers experiencing delays and others showing typical development.

Mixed Design Studies

A mixed design is used when it is not possible to randomly assign participants to groups. Instead, individuals are assigned to groups on the basis of a defined and differing variable, such as the presence of a cleft lip and/or palate. Mixed design studies are quasi-experimental, meaning that they do not carry the same high level of evidence as randomized control trials. However, this study design remains a valuable research tool because it allows researchers to examine whether correlations exist between group membership and a particular skill.

Broen, Devers, Doyle, Shirley, McCauley-Prouty and Moller (1998) conducted a mixed design to determine if the early cognitive and linguistic skills of children with NCL/P were delayed relative to their noncleft peers. Participants of this study included 28 toddlers with nonsyndromic cleft palate and a control group of 29 toddlers. Participant eligibility criteria was well specified. The participants were seen in three-month intervals between 9 and 30 months of age. At each session, the children were videotaped playing with their parents. Appropriate inter-rater reliability was reported for the transcription and scoring of the language samples, but no intra-rater reliability was listed. In order to measure the growth in each participants' vocabulary size, parents were instructed to write down each new word their child used between the ages of 12 and 24 months. Additionally, one scale from a gold standard developmental measure was administered at 24 months and two subscales from a different developmental measure were administered at 30 months to measure expressive and receptive language abilities.

Appropriate statistical analyses revealed significant differences in vocabulary size between the groups at 15, 18 and 21 months, receptive language measures at 24 months, and expressive and receptive language measures at 30 months of age. While toddlers with orofacial clefts scored lower than toddlers in the control group on expressive language measures at 24 months, no statistical difference was noted.

A limitation of this study is that the standardized assessment measures were not entirely appropriate as they did not provide a complete view of the children's language skills. Additionally, there are mixed reviews in the literature concerning the psychometric properties of one of the assessment measures. Lastly, although it was collected, there was no detailed information given concerning the children's vocabulary size at 24 months.

In summary, the results of this study provide somewhat suggestive evidence that the early linguistic skills of

toddlers with orofacial clefts are delayed relative to age-matched peers.

Scherer, Williams and Proctor-Williams (2008) implemented a mixed design in order to compare the early vocalization skills in children with NCL/P to their speech and vocabulary development at 30 months of age. By recruiting 13 participants with NCL/P and 13 noncleft toddlers, the authors were also able to investigate whether a difference existed between the two groups language abilities. Each participant was videotaped interacting with their caregivers at 6, 12 and 30 months of age. Appropriate inter- and intra-rater reliability was reported for the transcription and scoring of the language sample. Two gold standard expressive and receptive language measures were administered when the participants were 12 and 30 months of age. Appropriate statistical analyses revealed significantly lower scores for the NCL/P group than control group for babbling at 12 months and vocabulary and language at 30 months.

This study has several limitations including a small sample size, group differences in hearing at baseline and the use of one outdated language measure. Despite the limitations, this study provides compelling evidence that by 30 months of age, toddlers with NCL/P exhibit significant differences in early receptive and expressive language development when compared to noncleft peers.

Using a retrospective mixed design, **Hardin-Jones and Chapman (2014)** evaluated whether the early expressive lexicons of 37 toddlers with nonsyndromic cleft palate differed in size and lexical selectivity from those of 22 noncleft toddlers. Strict participant inclusion criteria were specified. The participants were videotaped interacting with their parents at 13, 17, 21 and 27 months of age. Two raters transcribed these interactions. Detailed transcription criteria and sufficient intra and inter-rater reliability was reported. To provide a measurement of the participants' vocabulary size, the parents completed a gold standard assessment during each of the home visits. Appropriate statistical analyses revealed significant differences in vocabulary sizes at 17, 21 and 27 months.

A limitation with respect to the study methodology is that the raters were not blinded to the participant's group assignment. Limitations also existed in terms of the group sizes and characteristics. There were more participants in the nonsyndromic cleft palate group than the noncleft group. While an attempt was made to match the two groups on relevant factors, the cleft group had a more significant history of ear infections and tympanostomy tube placement. Additionally, some toddlers in the cleft group were simultaneously receiving early language intervention to target expressive lexicon expansion. This may have reduced disparities in vocabulary size between the groups.

Overall, the study results provide suggestive evidence that at 17, 21 and 27 months of age, a significant difference in expressive lexicon size exists between the two groups that did not exist at 13 months of age.

Jocelyn, Penko and Rode (1996) utilized a mixed design in order to compare 16 children with NCL/P with 16 noncleft peers on measures of cognition, speech, language and audiologic status. The authors specified eligibility criteria and matched the two groups on relevant factors. A limitation of the study is that all of the participants were recruited from a single site. Two gold standard expressive and receptive language tests were administered at 12 and 24 months of age. At 24 months of age the children were videotaped interacting with their parents and a third gold standard expressive and receptive language test was completed. The authors did not report the transcription criteria for the language samples or inter/intra-rater reliability.

Appropriate statistical analyses revealed significantly lower expressive language scores for children in the NCL/P group at 12 and 24 months of age. There were mixed results concerning receptive language. Two of the assessment measures showed a significant difference between the two groups whereas the second measure did not.

A strength of the study is the use of multiple assessment measures as it provides a more complete picture of the child's language abilities. A limitation is that while the tests used were appropriate for the study question, they are now considered outdated tests.

In summary, the results of this study are highly suggestive that NCL/P differ significantly in their receptive and expressive language skills when compared to noncleft children.

Discussion

This review critically evaluated existing literature in order to determine whether the expressive and receptive language abilities of toddlers with NCL/P differ from their noncleft peers. Overall, the results from the selected studies provided highly suggestive evidence that toddlers with NCL/P score significantly lower on expressive language measures than age-matched peers. The results for receptive language were mixed.

Disagreement exists in the literature concerning whether the receptive language abilities of children with NCL/P fall in the deficit range. Two studies found that toddlers with NCL/P scored significantly lower than controls on receptive language measures (Broen et el., 1998; Scherer et al., 2008). However, one study did not report statistical differences between the two groups' performance on receptive language tests (Hardin-Jones & Chapman, 2011). One study reported mixed findings, with two of the employed assessments revealing significant differences between the groups while a third test found no significant difference in receptive language performance (Jocelyn et al., 1996). Despite the lack of consistent group differences, scores on receptive language measures were observed to be lower for children with NCL/P than controls in all studies. It is recommended that more research on the topic is completed in order to provide more conclusive evidence.

One trend that was noted in the research is that children with NCL/P were observed to have significantly smaller vocabulary sizes than age-matched controls. Three studies reported that at 12 months of age the NCL/P group and control group had similar vocabulary sizes, but by 15 months the NCL/P group had significantly smaller expressive lexicons (Broen et el., 1998; Scherer et al., 2008; Hardin-Jones & Chapman, 2014). The difference in vocabulary size persisted with age, with children with NCL/P showing significantly smaller vocabulary sizes at 30 months of age (Hardin-Jones & Chapman, 2014). These differences in vocabulary size were observed to increase with age, suggesting that children with NCL/P acquire words more slowly than controls (Jocelyn et al., 1996; Hardin-Jones & Chapman, 2014).

The current literature on the topic is characterized by small sample sizes. Although small sample sizes limit the generalizability of the findings to the larger NCL/P population, they are expected given the specific population being studied. There were several factors that made it difficult to compare the five articles. First, each study used different assessment measures to test the participant's language abilities. Secondly, the severity of the participants' cleft lip and palate varied within and between studies. Although each participant had a complete cleft of the palate, some participants also had accompanying unilateral or bilateral cleft lips.

Clinical Implications

Based on the findings of this critical review, it is recommended that a Speech-Language Pathologist regularly assess and monitor the expressive and receptive language skills of toddlers with NCL/P throughout development. By doing so, language intervention can be implemented at the earliest possible time. This is critical as early language intervention is integral to the prevention of associated problems in communication, literacy and cognition (Shonkoff & Phillips, 2001).

Future Research

Additional research is recommended to strengthen the current level of evidence. In future studies, the following recommendations should be considered:

I. Multi-site studies with larger sample sizes should be conducted in order to increase the generalization of the results to the larger NCL/P population;
II. Comprehensive expressive and receptive language assessment measures that are sensitive to detecting group differences in young children should be administered;
III. Strict participant inclusion criteria to ensure only toddlers with nonsyndromic cleft palate are included in the study.

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