Critical Review: How does age of palatal repair affect speech outcome in children with cleft palate?

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This critical review examines the speech outcome of children with cleft palate in relation to the timing of cleft palate repair. Overall, research suggests that the timing of palatal repair has some effect on the speech outcome, specifically that earlier repair appears more beneficial to speech development. Children in earlier repair groups demonstrated fewer symptoms of cleft speech such as nasality and misarticulation. The findings of this review have implications for further research and clinical practice in the field of speech-language pathology.

Introduction

The treatment objectives for cleft palate patients are normal speech, normal maxillofacial growth, and normal hearing (Rohrich, Love, Byrd & Johns, 2000). There is a longstanding controversy surrounding the relationship between these objectives and how the timing of cleft palate surgery affects the outcome of each. Generally, earlier cleft palate repair (prior to 24 months of age) has been deemed more advantageous to speech and hearing growth, while delayed closure (after 4 years of age) has been thought to improve maxillofacial growth (Rohrich, Love, Byrd & Johns, 2000). Timing is considered a critical factor in cleft palate treatment because children with cleft palate are potentially at a disadvantage during the prelinguistic phase of speech development due to the structural deviations associated with clefting (Ysunza et al, 1998).

Research in this domain has been difficult due to the high number of variables, such as physical variance of clefts, differences in surgical skill, lack of standard speech evaluation, and invariably the complexity of maturation, growth and development (Peterson-Falzone, 1996). It is likely due to these methodological and environmental factors that the optimal timing for palatal repair has not been scientifically proven (Leow & Lo, 2008).

As active members in an interdisciplinary team approach to cleft palate treatment, speech-language pathologists must be knowledgeable in current controversies surrounding approach to treatment, including the critical issue of timing of surgical repair. The evidence surrounding speech outcomes of children born with cleft palate should be critically examined and understood, so that the discerning clinician may appropriately contribute to the decision-making process. Although optimal timing for repair has not been clearly established, the statistical and descriptive evidence surrounding speech outcome is indicative of the need for speech pathology services as part of the cleft palate treatment team in general.

Objectives

The primary objective of this paper is to critically evaluate existing literature regarding the impact of timing of palatal repair on speech outcome in children with cleft palate. The secondary objective is to propose evidence-based practice recommendations for speech-language pathologists involved in cleft palate treatment.

Methods

Search Strategy

Computerized databases, including Scopus and Medline were searched using the following search strategy: (cleft palate repair) OR (timing of palate surgery) AND (speech) OR (speech outcomes).

The search was limited to articles written in or translated to English between 1985 and 2008. Articles were also located using references of reputable articles.

Selection Criteria

The studies that were selected for this critical review paper investigated the differences in speech skills of children whose cleft palates were surgically repaired earlier versus later in childhood, where timing of hard palate repair was the independent variable. No limits were set on the demographics of research participants or speech outcome measures.

Data Collection

Results of the literature search yielded the following types of articles congruent with the aforementioned selection criteria: randomized clinical trial (RCT) (1) and cohort study (5).

Results

Rohrich et al (1996) studied 44 randomly selected patients with a complete cleft palate treated at the Oxford Plastic Surgery Department between 1960 to 1969 in their retrospective cohort study. Patients were grouped based on age of hard palate repair (average 10.8 months in the early group, 48.6 months in the late group) and seen individually by a member of the Oxford Cleft Palate Study team for speech, maxillofacial, palatal and hearing assessments. The mean age of follow-up was 17 years of age in early group and 18.2 years of age in late group. Speech measures included: articulation, phonation, nasal emission, nasal resonance, intelligibility, and substitution patterns.

Significant differences in several speech measures of early and late closure groups were identified. The late closure group was found to have fewer incidences of normal nasal resonance ($p<0.001$), more hyponasality ($p<0.01$), and lower speech intelligibility ($p<0.02$). Phonation was found to be normal in both groups and no significant differences in nasal emission was noted. In regards to substitution patterns, both groups showed similar numbers of subjects with normal and midpalatal articulation patterns. The late closure group was observed to be the only subjects to use dentoalveolar patterns, while the early closure group demonstrated the velar pattern more frequently.

Ysunza et al (1998) used a randomized clinical trial design in their prospective study of speech outcome and maxillofacial growth in 76 patients with unilateral complete cleft palate. Subjects were randomly selected for the study group and subsequently randomly assigned to group for either early surgical repair (6 months of age) or late surgical repair (12 months of age) using the same surgical procedure. Subjects were followed until 48 months of age, at which time complete speech and maxillofacial evaluations were administered. The standardized speech evaluation included a clinical assessment of nasal resonance and articulation, with special consideration of compensatory articulation patterns.

Analysis of articulation development revealed that both groups performed below age expectations, however a Student's *t* test showed that the articulation scales of early repair group were significantly better than the late repair group ($p<0.05$). There was no significant difference in nasal resonance. However, only subjects in the late closure group with velopharyngeal insufficiency demonstrated compensatory articulation strategies.

A retrospective cohort study conducted by Kirschner et al (2000) compared the speech of 90 patients with unilateral cleft lip and palate who underwent palatal repair from 3 to 7 months of age (early repair group) and after 7 months of age (late repair group) between 1979 and 1992. A standardized speech evaluation, the Pittsburgh Weighted Values for Speech Symptoms Associated with Velopharyngeal Incompetence, was performed, including measures of nasal emission, facial grimace, nasality, phonation, and articulation. The mean ages of follow-up were 8.3 years (early repair group) and 8.7 years (late repair group).

Results indicated no significant differences between the groups in regard to resonance or nasal air emission ($p<0.05$). Only one person in each group demonstrated articulation errors related to velopharyngeal incompetence.

In their longitudinal cohort study, Lohmander et al (2006) report on the speech skills of 26 patients with complete unilateral cleft lip and palate operated on between 1996 and 1998. These patients were separated into two groups based on the timing of palatal repair: early repair group (38-52 months of age) and late repair group (64-89 months of age). Speech samples were collected at ages 3, 5, 7, and 10 years. At age 3, speech data were taken according to a specific and standardized protocol from patient records. Speech analyses at 5, 7, and 10 years of age consisted of a standardized clinical speech assessment and assessment by an external judge. This included the team speech-language pathologists' ratings of the following speech variables: hypernasality, weak pressure consonants, nasal air leakage, retracted oral articulation, and glottal articulation based on repetition of sentences and spontaneous speech. An external speech-language pathologist also made blinded judgments of velopharyngeal incompetence and articulation deficit based on audio recordings of the samples.

Results of longitudinal and descriptive analyses revealed no significant differences between groups at baseline or any of the follow-up ages. Only one speech variable, nasal air leakage, was affected by the group factor ($p<0.05$), where the late repair group showed more evidence of nasal air leakage. The factor of time appeared to have more significant impact on the variables of nasal air leakage, weak pressure consonants, and retracted oral articulation ($p<0.01$).

Holland et al (2005) performed a retrospective review of the speech and maxillary growth outcomes of patients with unilateral complete cleft lip and palate repaired between 1978 and 1988. The patients were separated based on surgical approach taken: early repair (single-stage at age 1) or late repair (soft palate repair at age 1, hard palate repair at age 7). The speech measures evaluated included perceptual velopharyngeal function assessment and standardized

assessment (Pittsburgh Weighted Values for Speech Symptoms Associated with Velopharyngeal Incompetence test). Speech symptoms were scored at each clinic visit using a standard format including ratings for nasal emission, nasality, phonation and articulation. The elements of velopharyngeal incompetence, articulation assessment (delayed, disordered, compensatory patterns), fistulas, and tonsil/adenoid presence were also collected. Groups were compared based on pre-operative speech score, follow-up speech score (average of scores at all follow-up visits at ages 3, 6, and 12 months, and 2, 3, 4, 5 and 6 years post-operatively), final speech score (score at 15 to 20 years of age), speech testing results, and the number of secondary procedures performed for speech.

Results of analyses revealed similar speech scores for each group pre-operatively. Average follow-up scores were found to be statistically different ($p<0.01$), showing more nasal emissions, facial grimacing, nasality, phonation problems and misarticulations in the delayed repair group. Mean final scores were also significantly different ($p<0.05$), where the delayed repair group showed worse overall speech outcome.

The retrospective cohort study performed by Haapanen and Rantala (1992) examined 108 patients with isolated cleft palate who attended follow-up examinations at age 3 between 1978 and 1988. These patients were divided into three groups based on their age at time of palatal repair: group 1 (mean age of 12.9 months), group 2 (mean age of 18.5 months) and group 3 (mean age of 22.1 months). Speech assessment was based on speech using picture stimulation (materials from a Finnish articulation test) as well as continuous speech samples. Speech data from clinical records were analyzed for hypernasality, nasal air emission, and misarticulations. Based on the presence of cleft palate speech characteristics, participants' quality of speech was classified as normal or practically normal, mildly impaired, or moderately or severely impaired.

The speech outcome for these groups revealed that group 3 demonstrated significantly more misarticulations, fewer participants with "normal" quality of speech, and more participants with "moderately/severely impaired" quality of speech as compared to the earlier repair groups ($p<0.001$).

Discussion

Subject Selection and Characteristics

There was some variance in the type of clefts seen in the samples across the studies analyzed. The samples of Holland et al (2007), Kirschner et al (2000), Ysunza et al (1998), and Lohmander et al (2006) included patients with complete unilateral cleft lip and palate. Ysunza et al (1998) in particular included patients with either a complete unilateral cleft lip and palate or unilateral cleft palate alone, but did not indicate the distribution of unilateral and bilateral clefts within the experimental groups. Haapanen and Rantala (1992) grouped patients with isolated cleft palate. Rohrich et al (1996) evaluated patients with complete unilateral or bilateral cleft palate, ensuring that a similar number of each (unilateral or bilateral) were in each experimental group. The distribution of males and females in the experimental groups of Rohrich et al (1996), Ysunza (1998), and Lohmander et al (2006) was similar. All other studies did not indicate the male to female ratio of participants. It is important that researchers specify the identifying characteristics of their sample, such that it is evident whether timing is the only difference between experimental groups. Any distinguishing characteristic differences (i.e. severity and type of cleft) reduce the possibility for drawing conclusions on the population as a whole.

Participants were accumulated from one specific hospital or clinic in each case, during a specific time period. Haapanen and Rantala (1992), Holland et al (2007), Ysunza (1998), and Kirschner et al (2000) outlined specific criteria for exclusion, including characteristics such as hearing loss, presence of fistulas, other syndromes, mental retardation, other medical problems, and insufficient data (incomplete medical history or failure to complete all follow-up sessions). Lohmander et al (2006) excluded participants who did not demonstrate speech difficulties at baseline from their between groups analyses. This type of exclusion potentially skews comparison between experimental groups, as it is not reported how many participants in either group were excluded for demonstrating no speech difficulties. Rohrich et al (1996) did not specify any exclusion criteria, reducing the possibility for accurate comparison with the studies whose exclusion criteria were described in depth. It is also important to consider that mixing results from typically developing children with those from syndromic or cognitively delayed patients will not facilitate the knowledge base necessary to make appropriate clinical decisions in these cases (Peterson-Falzone, 1996). On the other hand, while these criteria isolate the cleft as the sole concern, they do not allow for generalization to the wider population of individuals with cleft lip and/or palate who often present with concomitant anomalies or difficulties.

An important criterion not addressed in many of the studies was that of speech therapy. Haapanen and Rantala (1992) indicated that none of their participants had received speech therapy prior to their

follow up testing at age three. The only other researchers to mention speech therapy were Lohmander et al (2006), who indicated that some participants' medical history denoted attendance at speech therapy, while others' did not. This is a limitation of Lohmander et al (2006) and the remaining studies, as speech therapy could significantly impact the speech skills of any of the participants, regardless of age of palatal repair. Without considering the effect of speech therapy, the results of comparisons between early and late repair groups will not accurately reflect the influence of timing of palatal repair alone.

While the studies varied in their sample size, Haapanen and Rantala (1992), Holland et al (2007), Kirschner et al (2000), Rohrich et al (1996) and Ysunza et al (1998) included an adequate number of participants in order to make some general statements regarding the population (108, 82, 90, 44, and 76 participants, respectively) and have a higher degree of confidence in the studies' ability to detect differences between the groups. However, Lohmander et al (2006) presented the cases of only 26 patients (groups of 17 and 9). This low number of participants and uneven group distribution limits the generalizability of the researchers' findings and reduces the probability of detecting existing differences. This small sample size may also explain why Lohmander et al (2006) were unable to detect a difference between the speech skills of the early and late repair groups.

Methods

Each of the studies analyzed used a different set of speech measures to qualify the speech characteristics of the participants, as well as different standardized and subjective assessment measures. Characteristics common to all studies included articulation and nasal resonance. Phonation and substitution or compensatory articulation patterns were also common measures among some of the studies. The studies each utilized some measure to indicate degrees of these characteristics demonstrated by the participants, with the exception of Haapanen and Rantala (1992) who indicated presence or absence of characteristics only. In an effort to be more descriptive and compare the speech characteristics of participants more thoroughly, degrees or descriptors within each of the measures is an appropriate method for evaluation of speech outcome. A classified and detailed analysis of nasality and articulation may reflect compensatory speech strategies stimulated by any velopharyngeal inadequacy based on timing of palatal repair (Haapanen & Rantala, 1992).

The Lohmander et al (2006) study utilized both internal and external examiners, where only the external examiner was reported to be blinded. All members of the interdisciplinary team in the Rohrich et al (1996) study were blinded. These studies attempt to diminish some of the potential bias of the examiners. The randomized control trial conducted by Ysunza et al (1998) employed a double blind procedure, eliminating potential for bias on the part of the examiners as well as the participants. The remaining studies do not indicate whether a single or double blind procedure was used.

High interrater reliability was reported for Haapanen & Rantala (1992), Kirschner et al (2000), and Ysunza (1998), indicating good reliability of the results obtained by the speech assessments. The remaining studies did not indicate measures of interrater reliability. Interrater reliability is a critical component of these studies in order to establish the validity of the assessment results and conclusions drawn from them.

The validity of the speech measures used by each of these studies is an important consideration. Lohmander et al (2006) commented that their speech measure had high validity and was used in their treatment centre, but did not indicate what this measure was or give any proof of validity. Haapanen and Rantala (1992) did not use any standardized measures. The authors elicited samples informally and rated participants based on the speech sample and velopharyngeal function tests. Ysunza et al (1998) indicate that a standardized speech assessment was completed but do not indicate what the measure was. Rohrich et al (1996) utilized the Edinburgh articulation test, but also did not include indications of validity. Each of these researchers failed to appropriately indicate the validity of the measures used, diminishing the ability to compare the measures' efficacy and the results obtained. Holland et al (1996) and Kirschner et al (2000) both used the Pittsburgh Weighted Values for Speech Symptoms Associated with Velopharyngeal Incompetence test. Kirschner et al (2000) suggest that this Pittsburgh measure may not have been able to detect differences between the early and late repair groups in their acquisition of good speech, potentially accounting for this study's results indicating no significant differences between the groups.

Peterson-Falzone (1996) points out the importance of checking whether speech assessments were made by a speech language pathologist. In all studies, with the exception of Haapanen and Rantala (1992), the speech assessments were conducted by experienced speech-language pathologists. The participants in the Haapanen and Rantala (1992) study were assessed by the authors, who are

phoniatricians. This is a limitation of this study, as an appropriate assessment by a speech-language pathologist, including standardized measures, may have significantly altered the results.

The average ages of follow up for each of the studies varied. On one end, Haapanen & Rantala (1992) and Ysunza (1998) conducted their follow up evaluations at ages 3 and 4 years, respectively. Follow up at these young ages, while important to gather developmental data, do not reflect long term articulation difficulties. Articulation at this early age may also be attributed to typical developmental errors or substitution patterns, for which the researchers did not appear to account. On the other end, Kirschner et al (2000) and Rohrich et al (1996) followed up with their participants at the mean age of 8.3/8.7 (early/late group) and 17.5 years, respectively. These later follow up times take into account developmental changes after typical age of sound acquisition. Holland et al (2007) followed up with their participants at ages 2, 3, 4, 5, and 6, and Lohmander et al (2006) conducted follow ups at ages 3, 5, 7, and 10. These longitudinal studies, as well as the studies with later follow up ages, allow for observation of long term effects of cleft palate repair. However, maturation, speech therapy, exposure to speech and language in school, and other interventions must be taken into consideration when comparing speech shortly after surgery to speech several years later.

Recommendations

There is a sufficient body of evidence supporting the belief that earlier palatal repair is more beneficial for speech development. While the literature reviewed here demonstrates some limitations, the validity and reliability of the results as a whole have significant clinical implications. This review also influences future research in this area.

It is recommended that further research on this topic be completed to clarify and verify the relationship between timing of palatal repair and speech outcomes. In order to improve upon the evidence provided by the existing literature, it is recommended that future research take the following into account:

a) Clearly distinguishing experimental groups according to types (cleft lip and/or palate, unilateral or bilateral), severity of clefts, concomitant anomalies or difficulties, and receipt of speech therapy in order to increase possibility for accurate conclusions regarding the effect of timing of palatal repair alone.
b) Specific explanation of exclusion criteria, including identification of typically developing versus syndromic or cognitively delayed subjects, in order to increase generalizablity to this population.
c) Adequate sample sizes and distribution of participants into experimental groups that will facilitate appropriate conclusions for the population as a whole.
d) Use of suitable speech assessment measures that will allow an organized and descriptive examination of speech characteristics demonstrated as a result of velopharyngeal incompetence such that groups may be compared in greater detail. In addition to this, clearly identifying what these assessment measures are and indicating their validity.
e) Completion of speech assessments by a speech-language pathologist, clear identification of interrater reliability, and use of blinding procedures for examiners and subjects in order to increase the reliability of the assessments and the conclusions drawn from their results.
f) Consider mean age of follow-up, taking into account typical developmental errors when assessing younger children and considering the effects of maturation, speech therapy and other interventions when completing longitudinal analyses.

Clinical Implications

The results and conclusions discussed in this critical review have important clinical implications for speech-language pathologists. Despite the aforementioned limitations of these studies, the evidence presented here indicates a relationship between the timing of palatal repair and speech outcome. As suggested by Peterson-Falzone (1996), speech-language pathologists evaluating this literature should consider whether speech judgments were made by a speech-language pathologist and the nature of follow-up (i.e. longitudinal or one-time assessment in preschool years) in particular prior to establishing its significance.

It is essential that speech-language pathologists participate in the assessment and treatment of individuals with cleft palate with an understanding of the current literature related to speech outcomes. While the optimal timing of palatal closure remains unclear, the speech-language pathologist with a thorough understanding of this knowledge base will possess a higher degree of expertise concerning clinical cleft palate treatment, and advocate for the best available treatment to facilitate optimal speech development.

References

Haapanen, M., & Rantala, S. (1992). Correlation between the age at repair and speech outcome in patients with isolated cleft palate. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery, 26*(1), 71-78.

Holland, S., Gabbay, J. S., Heller, J. B., O'Hara, C., Hurwitz, D., Ford, M. D., et al. (2007). Delayed closure of the hard palate leads to speech problems and deleterious maxillary growth. *Plastic and Reconstructive Surgery, 119*(4), 1302-1310.

Kirschner, R. E., Randall, P., Wang, P., Jawad, A. F., Duran, M., Huang, K., et al. (2000). Cleft palate repair at 3 to 7 months of age. *Plastic and Reconstructive Surgery, 105*(6), 2127-2132.

Leow, A., & Lo, L. (2008). Palatoplasty: Evolution and controversies. *Chang Gung Medical Journal, 31*(4), 335-345.

Lohmander, A., Friede, H., Elander, A., Persson, C., & Lilja, J. (2006). Speech development in patients with unilateral cleft lip and palate treated with different delays in closure of the hard palate after early velar repair: A longitudinal perspective. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery, 40*(5), 267-274.

Peterson-Falzone, S. J. (1996). The relationship between timing of cleft palate surgery and speech outcome: What have we learned, and where do we stand in the 1990s? *Seminars in Orthodontics, 2*(3), 185-191.

Rohrich, R. J., Love, E. J., Byrd, H. S., & Johns, D. F. (2000). Optimal timing of cleft palate closure. *Plastic and Reconstructive Surgery, 106*(2), 413-421.

Rohrich, R. J., Rowsell, A. R., Johns, D. F., Drury, M. A., Grieg, G., Watson, D. J., et al. (1996). Timing of hard palatal closure: A critical long-term analysis. *Plastic and Reconstructive Surgery, 98*(2), 236-246.

Ysunza, A., Pamplona, M. C., Mendoza, M., García-Velasco, M., Aguilar, M. P., & Guerrero, M. E. (1998). Speech outcome and maxillary growth in patients with unilateral complete cleft lip/palate operated on at 6 versus 12 months of age. *Plastic and Reconstructive Surgery, 102*(3), 675-679.