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Speech outcomes at age 5 and 10 years in unilateral cleft lip and palate after one-stage palatal repair with minimal incision technique – A longitudinal perspective☆

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ABSTRACT

Objectives: To investigate speech outcomes in 5- and 10-year-old children with unilateral cleft lip and palate (UCLP) treated according to minimal incision technique (MIT) – a one-stage palatal method.
Methods: A retrospective, longitudinal cohort study of a consecutive series of 69 patients born with UCLP, treated with MIT (mean age 13 months) was included. Forty-two children (43%) received a velopharyngeal flap; 12 before 5 years and another 18 before 10 years of age. Cleft speech variables were rated from standardized audio recordings at 5 and 10 years of age, independently by three experienced, external speech-language pathologists, blinded to the material. The prevalences of cleft speech characteristics were determined, and inter- and intra-rater agreement calculated.
Results: More than mild hypernasality, weak pressure consonants and perceived incompetent velopharyngeal function were present in 19–22% of the children at 5 years, but improved to less than 5% at 10 years. However, audible nasal air leakage, prevalent in 23% at 5 years, did not improve by age 10. Thirty percent had frequent or almost always persistent compensatory articulation at 5 years, and 6% at age 10. The general impression of speech improved markedly, from 57% giving a normal impression at 5 years to 89% at 10 years. A high prevalence of distorted/s/was found at both 5 and 10 years of age.
Conclusions: A high occurrence of speech deviances at 5 years of age after MIT was markedly reduced at 10 years in this study of children with unilateral cleft lip and palate. The high pharyngeal flap rate presumably accounted for the positive speech development.

1. Introduction

The necessity of expanding reliable knowledge regarding speech development and surgical methods is essential in providing care for children born with cleft lip and palate. Although this has been a matter of course for decades, cleft palate care has relied on studies with limited scientific basis for intervention [1]. There is still no consensus on either the timing or techniques of palatal surgery and knowledge about speech outcome in children born with cleft lip and palate is still sparse: "What to do and when to do it remain constant concerns of teams and surgeons" [2, p. 149].

1.1. Surgical timing and technique

The concerns regarding which surgical method provides the best results are related to the goal of achieving both adequate midface growth and normal speech development [2]. Early closure of the palatal cleft is recommended for improved speech and over the years palatal surgery has tended to be performed at younger

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ages. The common practice today is to perform one-stage palate repair at between 6 and 18 months of age [1, p. 56]. With a one-stage method both the hard and soft palate are closed in a single session. An alternative opinion advocates a two-stage protocol with an early repair of the soft palate, usually carried out between 3 and 9 months [3], followed by hard palate closure in later childhood. The purpose of a delayed palatal repair is to minimize maxillary growth impairment [4]. Another alternative option for the two-stage palatoplasty is to close the anterior hard palate with a vomerine flap at the time of the lip and nose surgery and then close the soft palate, usually at around 6 months [5].

Not only do the staging and timing of palatal surgery vary but also the surgical techniques practiced, particularly for soft palate repair. In order to minimize scarring a minimal incision palatoplasty was described Mendoza et al. [6] and later by Kriens [7]. The intention of the procedure was "to allow good muscular reorientation as well as elongation of the soft palate with minimal morbidity and scarring" [6, p. 199]. A technique with a more radical muscle reconstruction of the soft palate is often recommended today, claiming better speech results [8,9].

1.2. Speech outcomes between 5 and 12 years of age

The ages when children are about to enter school and adolescence are the most commonly reported outcome ages in the literature [1]. The most distinctive speech disorders associated with cleft lip and palate are deviant consonant production, hypernasality, and audible nasal air leakage. These are usually reported as cross-sectional speech outcomes even though the terminology and procedures for assessment vary. Sell et al. [10] in their national prospective study, reported on speech outcomes at 5 years of age in 238 children born with UCLP and treated at different centers in the United Kingdom. The surgical method used for repair of the palate varied among the cleft centers. The results for these children showed that 34% had at least one serious error in consonant production, 29% some degree of hypernasality and less than 20% had entirely normal intelligibility. Poorer results regarding hypernasality was presented by Pigott et al. [11] in their comparison of speech outcomes at 5 years of age in 66 children after three different surgical methods for one-stage palatal repair. They found that around 45% of the 5-year olds had hypernasality, with no significant difference related to surgical method. The occurrence of cleft-related articulation errors was also high but varied between the surgical methods (32–78%). Pulkkinen et al. [12] included 30 children with UCLP and speech was evaluated at different ages including 6 years of age showing that hypernasality was present in 41.5%. Compensatory articulations were present in 12% of the children at the same age. Timmons et al. [13] reported better speech outcomes in 17 children with UCLP including 5 years of age (mean: 7.8 years). None of the children had severe hypernasality, and 23.5% had mild consistent hypernasality. Articulation errors were found in 53% of the children but were comprised only of anterior oral errors.

While Sell et al. [10] who also investigated a large group of 12-year olds, found that 19% had speech that was different enough to provoke comments, speech has been reported to improve for most children at around that age [14–16]. In these studies of children treated according to different surgical procedures, the majority had normal speech by the age of 10–12 years. However, two recent small-sample cross-sectional studies of speech at 10 years of age in children born with UCLP reported poorer outcomes on hypernasality and also on articulation; one after a two-stage [17] and one after a one-stage [18] palatal surgical procedure. The results, showing that between 34% and 46% of the 10 years old children in these two studies had speech problems.

Since treatment of hypernasality is velopharyngeal surgery, the need for secondary surgery is sometimes used as an indicator of the success rate with the primary palatal operation. The frequency of velopharyngeal flaps in children with UCLP in the studies above varied between 10% and 20% with no clear difference related to the performed primary palatal surgical procedure [10,15–17,19].

In order to study the development of speech longitudinal studies are needed. To date, most longitudinal studies have been performed after two-stage palatoplasty in children with UCLP, two including 5- and 10-year olds [16,20]. In these, the external expert listeners found that 30–40% of the children had oral consonant errors at 5 years in these studies. The corresponding figure at 10 years of age was approximately 10%. The oral articulation errors for the children in the studies were mainly retracted oral articulation, that is anterior/alveolar pressure consonants produced in palatal/velar position. The degree of hypernasality varied slightly in the studies, but at 5 years of age, when the cleft in the hard palate was as yet unoperated, around 30% of the children with UCLP were judged to have moderate to severe hypernasality. Lohmander et al. [16] found that 6% of the children in their study were judged as having more than mild hypernasality at 10 years of age (after hard palate closure). In that study, 11% of the children had received a pharyngeal flap. A gradual improvement in speech over time was also reported by Havstam et al. [20], but no information on pharyngeal flap rate was given. Whereas mild or moderate impairment were most prevalent at 5 years of age in their study, a clear majority had normalized articulation and intelligibility at 10 years. However, almost 50% of the children were assessed as having mildly or moderately impaired overall aspects of speech, such as perceived velopharyngeal function and a mild deviant general impression of speech at 10 years. None of the studies provided detailed information on speech therapy, consequently the influence of speech therapy was not clarified.

The assumption that the number of children with speech problems considered in need of speech therapy will decrease with improved surgical treatment programs has until now not been proven right, and the empirical evidence regarding the impact of speech therapy for children with cleft palate is limited in both quantity and design [2]. In a recent systematic review, Besell et al. [21] found no evidence to support any specific speech therapy intervention. Nevertheless, several studies have revealed that many children receive massive speech intervention, either a combination of surgery and therapy or one of them, depending on the speech deviances [10,12,17].

1.3. Purpose of the study

Knowledge about speech development in children born with cleft lip and palate is still sparse at the important ages when children are about to enter school and adolescence, which are both vulnerable periods in a person's life. The overall aim of this study was therefore to investigate longitudinal speech outcome at these relevant ages in a cohort of children born with UCLP treated with a one-stage palatal procedure.

The specific research questions were:

1. What is the prevalence of typical cleft speech characteristics at 5 years of age after one-stage palatal repair?
2. How do cleft speech characteristics compare from 5 to 10 years of age after one-stage palatal repair?
3. Is there an association between age at primary palatal surgery or amount of speech therapy administered, on speech outcome?

Ethical approval was given by the Ethical Regional Committee in Stockholm (Dnr 2011/2065-31/3).