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The effect of age at surgery and compensatory articulation on speech outcome in submucous cleft palate patients treated with double-opposing Z-plasty: A 10-year experience

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Summary *Background:* Submucous cleft palate is a congenital deformity that may present as velopharyngeal insufficiency. The degree of anatomical abnormality varies widely among patients and does not predict severity of symptom. We present our 10-year experience treating submucous cleft palate patients with double opposing z-plasty and explore the effect of age at surgery and compensatory articulation on speech.
Methods: Preoperative assessment included intraoral examination, nasoendoscopy, and perceptual speech evaluation. Age, gap size, and severity of hypernasality were assessed to determine the timing and type of surgery. A retrospective study of 74 submucous cleft palate patients undergoing double-opposing z-plasty from 2005 to 2016 by a single surgeon (Baek RM) was conducted. Double opposing z-plasty was modified to fully release all abnormal insertions of the *levator veli palatini* muscle. Postoperative velopharyngeal function was measured and statistical analyses were performed.
Results: The ages of patients at surgery ranged from 11 months to 19 years. Postoperatively 87 percent of the patients achieved velopharyngeal competency and 13 percent had remaining mild hypernasality. Age was a significant factor in predicting speech outcome, and patients receiving surgery prior to the age of 5.5 years had a higher normalization rate. Compensatory

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articulation did not have an impact on postoperative hypernasality. There were no cases of postoperative hyponasality or airway problems.
Conclusion: Satisfactory speech outcome was achieved with the authors' protocol and modification of the double-opposing z-plasty. A comprehensive assessment of patient age, intraoral findings, severity of hypernasality, and gap size, is necessary for proper treatment of submucous cleft palate patients.

Introduction

Submucous cleft palate (SMCP) is a congenital deformity that is characterized by an abnormal soft palate, which can present as velopharyngeal insufficiency. The diagnosis of SMCP is made based on the clinical triad of anatomical features defined by Calnan in 1954, which consists of 1) a bifid uvula, 2) a notch in the hard palate posterior end, and 3) zona pellucida in the midline of the soft palate due to muscular diastasis.[1] An occult form of SMCP without the three overt signs also exists, and in such cases, there is a muscular defect in the soft palate that is only evident on nasoendoscopy.[2] SMCP and occult SMCP are believed to be variations along the spectrum of severity of muscular malformation and occult SMCP is thought to represent 10—20 percent of the SMCP phenotype.[3,4] The exact prevalence of SMCP in the general population is unknown, but Weatherley-White (1972) and colleagues found that 9 out of 10,836 (0.08 percent) school-aged children had the triad of SMCP, and Garcia-Velasco (1988) screened 6000 children based on presence of two of the cardinal signs and yielded a prevalence of 0.02 percent.[5,6]

SMCP patients may present with chronic hearing problems, feeding difficulties, hypernasal speech or any combination of the three. However, the anatomical abnormalities seen in SMCP do not necessarily produce symptoms. Weatherley-White (1972) reported five to ten percent occurrence of velopharyngeal dysfunction in individuals with SMCP based on a school survey.[5] Garcia Velasco (1988) found velopharyngeal insufficiency in 53 percent of SMCP patients in a referral-based study.[6] Once the diagnosis of SMCP is made, the speech of the child is the major factor that determines if surgery will be necessary. Speech evaluation for the presence of hypernasality is feasible for children around three years of age when they produce connected speech. SMCP with velopharyngeal insufficiency requires surgical correction, while asymptomatic SMCP requires no surgery.

A few studies have investigated the relationship between age at surgery and speech outcome. Porterfield and Trabue (1965) recommended repair at 16—18 months of age to avoid acquisition of undesirable speech habits that persist post-surgery.[7] Abyholm (1976) reported superior speech outcome in patients who underwent repair using a combination of von Langenbeck palatoplasty and pharyngeal flap or pharyngeal flap alone prior to age 7.[8] Pensler and Bauer (1988) emphasized the importance of early diagnosis and achieved better speech results in patients operated on prior to age 2.[9] To the authors' knowledge, existing studies regarding the effect of age at surgery and speech outcomes in SMCP patients have reported findings that are not consistent with one another, and thereby a guideline has not yet been established for the treatment of SMCP. Also, the impact of compensatory articulation errors prior to surgery on postoperative speech results has not yet been assessed in other studies.

The aim of this study was to describe our 10 years of experience diagnosing and treating patients with SMCP using modified double-opposing z-plasty and examine the effect of age at surgery and the presence of compensatory articulation errors prior to surgery, on speech.

Methods

This study was approved by the institutional review board of Seoul National University Bundang Hospital (IRB no. B-1607-354-112).

The current treatment protocol for SMCP at Seoul National University Bundang Hospital varies depending on patient age. If a patient is diagnosed before beginning to produce connected speech, the child is closely observed to see if he or she develops hypernasality. However, if the child has the apparent triad of signs and presents with feeding problems such as reflux of oral contents into the nose, along with recurrent otitis media with effusion, surgery is considered.

When the child can produce connected speech, we perform perceptual speech evaluation. Patients with velopharyngeal insufficiency on perceptual speech assessment undergo nasoendoscopy if cooperative. The type of surgical plan is determined based on the patient's age, velopharyngeal gap size, and severity of hypernasality. If there is a pinhole-sized gap with intermittent hypernasality, the patient is started on a trial of speech therapy and surgery is offered only in case of no improvement. Double opposing z-plasty is indicated when at least two of the following criteria are fulfilled; age $\leq$ 7 years; gap $\leq$ intermediate; hypernasality $\leq$ moderate. When the patient is not cooperative with endoscopy, the palate is examined for signs of obvious diastasis of levator veli palatini muscle. A zona pellucida or visible anteriorly-oriented palatal bundles indicates palatal muscle diastasis.[10] On the other hand, if intraoral findings are ambiguous, the patient is asked to return to the clinic when he or she is old enough to undergo nasoendoscopy.