Speech Outcomes of 10-year-old Children after Early Palatoplasty Using Presurgical Orthodontics at 6 Months of Age

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Objective: We aimed to assess whether patients who underwent early palatoplasty have normal speech.
Methods: 19 patients with unilateral cleft lip and palate were enrolled in this study. At 6 months of age, we performed simultaneous lip, maxilla, and palate repair using presurgical orthodontics. Speech development was assessed by evaluating velopharyngeal function (VPF) and development of articulation for 10 years.
Results: No articulation disorders were observed after 4 years of age. Although palatalized articulation was evidently temporary in 3 cases before 4 years of age, all patients recovered without any speech training. Normal VPF rates were as follows: at 4 and 7 years of age 78.9% (n = 15), 10 years of age 73.7% (n = 4). 10 patients temporarily presented with mild VPI after 5 years of age although they had a normal VPF until 4 years of age.
Conclusion: Early palatoplasty after narrowing the cleft palate using presurgical orthodontics is beneficial for development of articulation. The rate of normal VPF did not decrease over the years.

Key words: cleft palate, early palatoplasty, presurgical orthodontics, long term, speech

INTRODUCTION

Previous studies have reported normal speech outcomes after early palatoplasty [1–9]. Authors have claimed that the early reconstruction of normal anatomical structures of the palate contributes in achieving normal speech.

However, early palatoplasty had not been standardized because surgery of the immature palate of infants less than 1 year of age causes severe disturbances in maxillary growth [10, 11]. Therefore, we performed a less-invasive surgery after narrowing the cleft palate using presurgical orthodontics in children with unilateral cleft lip and palate (UCLP). This less-invasive surgery may circumvent maxillary growth disturbances and result in both normal speech and maxillary growth with palatoplasty at an early age of 6 months. In this study, we aimed to assess whether pediatric patients who underwent palate closure at 6 months of age have normal speech. For this purpose, we monitored and evaluated the speech outcomes of our patients for 10 years.

MATERIALS AND METHODS

Subjects

Totally, 30 pediatric patients from the plastic surgery department of Tokai University Hospital underwent the procedure between 2001 and 2008. Of these, 19 patients aged more than 10 years were enrolled in this study. Patients with mental retardation or anomaly syndromes were excluded. This research was performed with parental consent and approval from the judging committee of clinical research (registration number: 18R-102). The investigation conforms with the principles outlined in the Declaration of Helsinki.

Treatment protocol

At 2 months of age, patients were fitted with a Latham's orthodontic appliance [12]. Fitting was performed under local anesthesia. Alignment of the alveolar ridge and narrowing of the alveolar cleft were achieved in approximately 1 month using the same appliance. Subsequently, the patients were fitted with a novel palatal orthodontic appliance under general anesthesia at 5 months of age. The posterior part of the cleft was narrowed for approximately 1 month. At 6 months of age, simultaneous lip, maxilla, and palate repair was performed. Millard gingivoperiosteoplasty [12] was performed for alveoplasty.

Furlow's palatoplasty [13] was used, and the palate and alveolar ridge were connected and reconstructed. Lip repair was performed using Millard's rotation-advancement method with small triangular flap [14, 15]. Hospitalization period was 1 week.

Assessment

Speech development was assessed by evaluating velopharyngeal function (VPF) and the presence of articulation disorders. Regarding articulation disorders, we examined the presence or absence of glottal and palatalized and lateralized articulation. VPF outcomes were evaluated according to 4 grades: normal VPF and mild, moderate, and severe velopharyngeal insuf-

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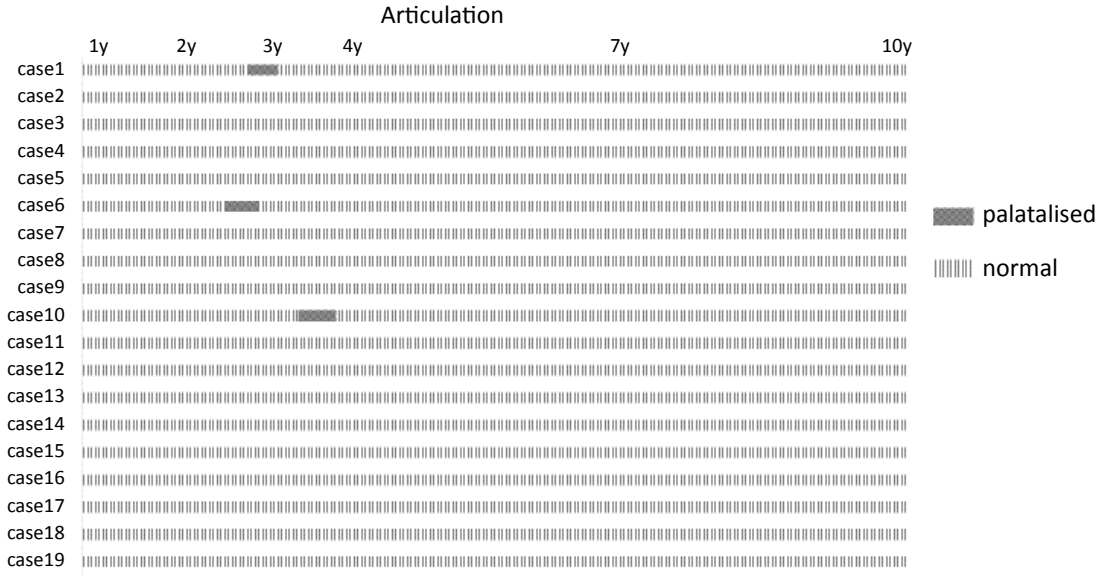

Fig. 1 Development of articulation in each patient. Although palatalized articulation was evidently temporary in cases 1, 6, and 10 at 2 or 3 years of age, all patients recovered without any speech training. All patients developed normal articulation until 4 years of age.

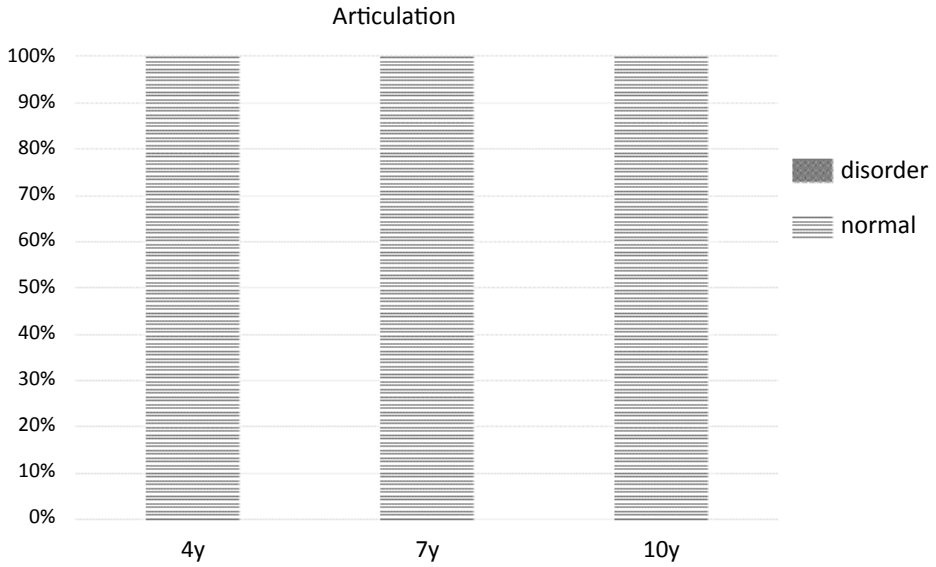

Fig. 2 At 4 and 7 and 10 years of age, 19 patients (100%) showed normal articulation. No articulation disorders were observed after 4 years of age.

ficiency (VPI). Speech was assessed by a senior speech pathologist using the Japanese standard test [16] established by the Committee on Cleft Palate Speech of the Japanese Association of Communication Disorders at 3-month intervals in patients up to 4 years of age and at 6-month intervals in patients more than 10 years of age.

RESULTS

Articulation

All patients developed normal articulation until 4 years of age. Although palatalized articulation was evidently temporary in cases 1, 6, and 10 at 2 or 3 years of age, all patients recovered without any speech training (Fig. 1).

Fistula closure was performed in case 10 at 1 year of age due to anterior palatal fistula. The patient showed palatalized articulation at 3 years of age after surgery; however, the patient also recovered without speech training. At 7 and 10 years of age, 19 patients (100%) showed normal articulation. No articulation disorders were observed after 4 years of age (Fig. 2).

VPF

(Fig. 3) shows the progression of VPF in each patient. Additionally, 10 patients temporarily presented with mild VPI after 5 years of age although they had a normal VPF until 4 years of age. In 7 patients, VPI improved spontaneously; however, it did not improve in the other patients. At 4 years of age, the normal VPF rate was 78.9% (n = 15), and mild VPI rate was 21.1% (n = 4). At 7 years of age, the normal VPF rate was 78.9% (n = 15). The normal VPF rate in the patients was 73.7% (n = 14) at 10 years of age. Further, 4 patients (21.1%) showed mild VPI, and 1 patient (5.3%) presented with moderate VPI (Fig. 4).

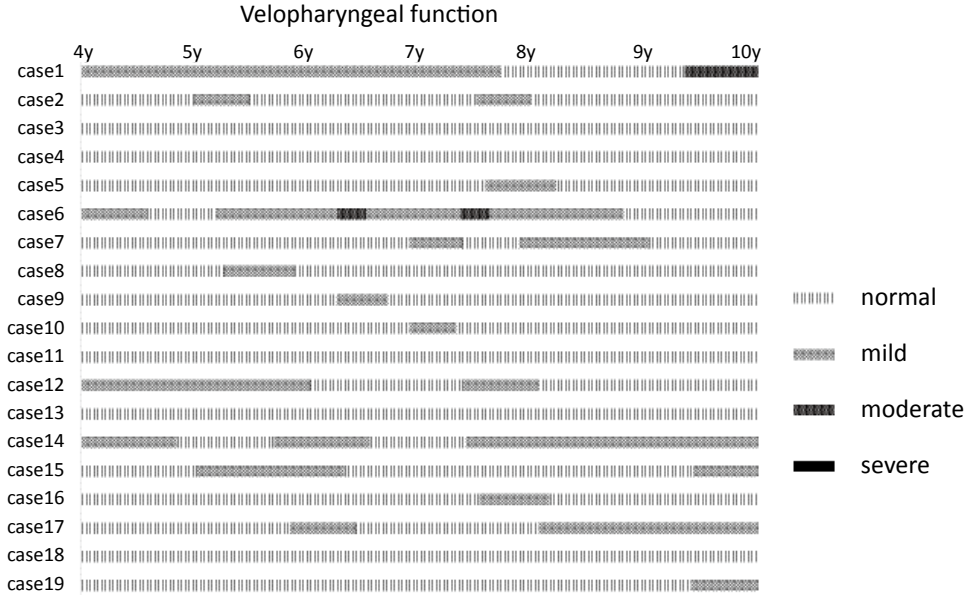

Fig. 3 Progression of VPF in each patient. 10 patients temporarily presented with mild VPI after 5 years of age although they had a normal VPF until 4 years of age. In 7 patients, VPI improved spontaneously; however, it did not improve in the other patients (cases 15, 17, 19).

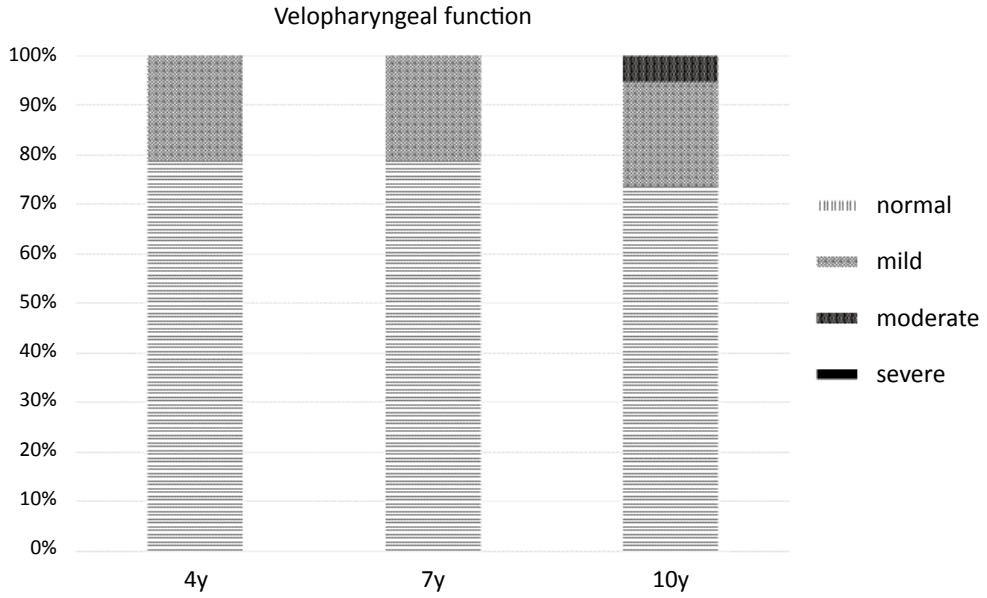

Fig. 4 At 4 and 7 years of age, the normal VPF rate was 78.9% (n = 15) and mild VPI rate was 21.1% (n = 4). At 10 years of age, one patient (5.3%) presented with moderate VPI.

DISCUSSION

Although there were variations in surgical procedure, timing of palatoplasty, and evaluation methods for speech, several authors have claimed that early palatoplasty results in better speech outcomes [1–9].

Articulation

In our patients, no articulation disorders were observed after 4 years of age. Some authors have reported that the timing of palatoplasty is associated with a lower incidence of articulation disorders (Table 1). Ysunza [1] and Dorf [2] have reported that the frequency of compensatory articulations developed significantly less in the early repair group than in the late repair group. A study by Randag [3] also showed a significant difference in articulation; the mean number of correctly produced initial consonants was higher in early one-stage palatoplasty.

Speech results following late primary palate closure (over 8 years of age) were reported by Murthy [17] and Bruneel [18] (Table 1). In late repair groups, the incidence of articulation disorder was remarkably higher than those of early repair groups of other studies. This suggested that delayed palatal repair was insufficient to eliminate articulation disorders.

As the reason for this, some authors have demonstrated as follows. Approximately 50% of infants exhibit canonical babbling by 9 months of age [19]. Thus, surgery at 6 months of age would ensure that palatal repair is performed prior to that time. A study by Kaplan E.N. [4] showed that the delay in palate

Table 1 Overview of studies reporting speech outcomes after early or late palatoplasty.

Author	Early repair				Late repair			
	Palatal closure: age and surgical technique	Age at speech assessment	Rate of normal articulation	Rate of normal VPF	Palatal closure: age and surgical technique	Age at speech assessment	Rate of normal articulation	Rate of normal VPF
Ysunza [1] (1998)	6m Minimal incisions palate-pharyngo-plasty	4y	100%	83%	12m Minimal incisions palate-pharyngo-plasty	4y	88%	81%
Dorf [2] (1982)	5–12m von Langenbeck	18–30m	83%	-	12–17m	18–30m	13%	-
Randag [3] (2014)	10.8m von Langenbeck or two flap method	2.5y	(9.71)*	73.9%	10.8m: soft palate 18.2m: hard palate Two-stage	2.5y	(6.96)*	66.7%
Dzierzbicka [6] (2012)	6–13m	10y	96.4%	89.2%				
Kirschner [20] (1998)	< 6m 6–12m	5y	-	80% 78%	12–18m > 18	5y	-	65% 84%
Jackson [21] (2013)	< 6m 6–12m Modified Furlow	5y	83.3% 95.1%	91.5% 91.7%	12–18m > 18 Modified Furlow	5y	97.7% 99%	89.6% 80%
Murthy [17] (2010)					> 10y Two flaps technique	10–35y	0%	0%
Bruneel [18] (2017)	Non cleft	8–29y	73%	100%	> 8y Sommerlad technique	18y10m	13%	20%
Park [22] (2000)					13–35m Push-back palatoplasty	4y > 10y	57.1% 83.9%	76.8%** 92.9%**
Hanai (This study)	6m Modified Furlow	4y 7y 10y	100% 100% 100%	78.9% 78.9% 73.7%			-	

* The number of correctly produced initial consonants is shown in parentheses.
** The number includes the patients having pharyngeal flap surgery.

repair induced abnormal compensatory habit patterns until 8–9 months of age and then remained after surgery because neuromuscular control and integration into the neurological system occur at an optimal time. Moreover, the repaired palate, like other tissues, has limited mobility because of postoperative edema and scarring for 3–6 months. Therefore, palatoplasty should be performed several months prior to normal speech onset to allow for scar maturation.

VPF

Previous reports have shown that the normal VPF rates were approximately 70%–90% (Table 1). The VPF result of our series was acceptable. Early reconstruction of the muscle sling of the soft palate contributes to a normal VPF [5].

Some authors have reported that the timing of palatoplasty is not associated with VPF [1, 20, 21]. Kirschner [20] examined VPF in a large series of Furlow's palatoplasties performed at a single center and reported that the age at palatoplasty had no significant effect on normal VPF rates. Jackson [21] evaluated retrospectively the speech outcomes at 5 years of age after a modified Furlow's palatoplasty in 559 pediatric patients at a single institution. He concluded that the age at repair was significantly associated only with a higher incidence of compensatory articulation errors. Resonance and nasal emission were not affected by age at repair.

In fact, our result of normal VPF rate was not excellent despite of early repair. However, to adapt to life in society, development of normal articulation is more important for children with cleft palate than obtaining normal velopharyngeal function.

Necessity of long-term follow-up

In our research, the rate of normal VPF did not decrease at 4–10 years of age. However, some authors have claimed that VPF is likely to decline over the years [22–25]. The causes for this condition included the following reasons: regression of adenoids and expansion of pharyngeal cavity caused by growth [23–28]. A study by Park [22] showed that 32.1% of patients with UCLP who underwent Push-back palatoplasty have deteriorated VPF at 4–10 years of age. On the other hand, the rate of articulation disorders improved from 4 (42.9%) to 10 (16.1%) years of age owing to the treatment of VPI and speech therapy. Therefore, Park concluded that speech outcome should be assessed longitudinally to better understand the changes in VPF and articulation disorders, and the final speech assessment should be carried out after 10 years of age.

In many previous reports, the age of evaluation was too young. Few authors have attempted to document the changes in speech outcome after early repair following over 10 years of age [6, 7]. In 2012, Dzierzbicka

[6] reported good speech result at 10 years of age after early simultaneous lip and palate repair.

Accurately evaluating the speech outcomes of such young child is not easy. Though factors of aged decline in VPF are generally considered regress of adenoids and the expansion of pharyngeal cavity with growth. However, it may also depend on the evaluation. There is a possibility of inaccurate evaluation at young age in previous studies.

Prevention of maxillary growth disturbance

Occlusion and facial growth should also be evaluated along with speech to confirm the superiority of early palatoplasty. Previous early palatoplasty has not been generalized due to maxillary growth disturbance. Early repair will never be accepted unless maxillary growth is considered. We had already investigated such countermeasures. Using an orthodontic appliance narrowed the cleft in the palate and allowed the use of less-invasive palatoplasty because of minimal lateral tension during palatoplasty. In this way we could accelerate timing of palate repair. When an invasive surgical procedure is not performed, it reduces the effect of maxillary growth due to minimal postoperative contracture. We propose to report the outcomes of maxillary growth in the next study after the patients grow.

CONCLUSION

The speech outcome of 19 patients with UCLP who underwent simultaneous lip and palate repair at 6 months of age was evaluated for 10 years. Early repair using presurgical orthodontics is beneficial for articulation. However, some patients temporarily presented with mild VPI after 5 years of age although they had a normal VPF until 4 years of age, and the rate of normal VPF did not decrease over the years.

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