The Effect of Preoperative Use of an Orthopedic Plate on Articulatory Function in Children With Cleft Lip and Palate

Keiko Suzuki, Ph.D., Yasuharu Yamazaki, D.D.S., Ph.D., Kouichiro Sezaki, M.D., Ph.D., Nobuaki Nakakita, M.D., Ph.D.

Objective: **To evaluate the effect of preoperative use of an orthopedic plate (OP) on postoperative articulatory function in children with cleft lip and palate.**

Subjects: **The subjects had complete unilateral or bilateral cleft lip and palate and were scheduled for a one-stage palatoplasty.**

Main Outcome Measures: **Tongue movements during sucking were analyzed by ultrasonography. Postoperative articulatory behavior was also assessed at 5 years 4 months of age.**

Results: **There was an excessive downward excursion of the rear portion of the tongue during sucking regardless of the use or nonuse of the OP. This indicated that infants with cleft palate could not create negative pressure in the oral cavity, even with the OP. However, the OP appeared effective for preventing irregular movements of the tongue during sucking. The proportion of subjects obtaining excellent articulation was significantly higher in the group using the OP until palatoplasty than in the group who did not continue using the OP. The proportion of subjects with disturbed articulatory function in the latter group was comparable with that in the control group, who never used the OP.**

Conclusions: **Continuous use of the OP up to the time of palatoplasty appeared to be effective for the postoperative articulatory function in children with complete cleft lip and palate. Inhibiting irregular movements of the tongue, the OP might assist in preventing "palatalized articulation."**

KEY WORDS: *articulatory function, cleft lip and palate, orthopedic plate, tongue movement, ultrasonography*

At Kitasato University Hospital, the use of an orthopedic plate (OP) was started in 1994 for the treatment of children with cleft lip and palate. The OP occludes the cleft only at the level of the hard palate (Fig. 1). Unlike the Zurich approach (Hotz et al., 1978, 1986), which uses a two-stage palatoplasty, we applied the plate in those children with cleft lip and palate who were scheduled for a one-stage palatoplasty by using the mucosal grafts and flaps method (Perko, 1974; Kamiishi, 1985; Torikai, 1995) or modified Furlow method (Torikai et al., 1994). These methods appeared to be harmless for jaw growth even if performed before the age of 2 years because the periosteum of the hard palate was well preserved (Torikai et al., 1997; Udagawa, 2001).

Dr. Suzuki is Speech-Language-Hearing Therapist and Lecturer of Speech Therapy Course, Kitasato University School of Allied Health Sciences, Kanagowa, Japan. Drs. Yamazaki and Sezaki are Lecturers and Dr. Nakakita is Associate Professor in the Department of Plastic Surgery, Kitasato University School of Medicine, Kanagowa, Japan.

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Address correspondence to: Keiko Suzuki, 1-15-1 Kitasato Sagamihara-shi Kanagawa, Japan 228-8555. E-mail urawa@ahs.kitasato-u.ac.jp.

Although the OP has been widely used as a preoperative treatment for children with cleft lip and palate, its rationale and effect are still under discussion (Winters and Hurwitz, 1995; Severens et al., 1998). There have been a considerable number of reports on articulatory development of children with cleft palate who used the OP preoperatively. Dorf et al. (1985) reported a 3-year study of 11 children who underwent early prosthetic obturation. Those who failed to wear the prosthesis and had late surgical palate repair demonstrated a predominance of compensatory articulations. However, one of two babies who used the prosthesis until palatoplasty demonstrated normal speech development. Konst et al. (1999) described the effect of the use of the OP on consonant development in babbling. They found the enhanced production of alveolar consonants in babbling at 1 year of age, contrary to Hardin-Jones et al. (2002), who did not find significant effects from the use of the OP on the place of consonant production in babbling. Konst et al. (2000) described a beneficial effect of using the OP on judged intelligibility in children between the age of 2 and 3 years. Most of these reports simply dealt with the children's early speech development, and no comprehensive observation has been made on its long-term effect on postoperative articulatory function.

FIGURE 1 OP of Kitasato team.

For the past 3 decades in Japan, 40% to 50% of the children with cleft palate developed abnormal articulation regardless of the surgical methods of palatoplasty, except for the two-stage operation (Ainoda et al., 1985; Okazaki et al., 1985; Yoshimasu et al., 1986; Suzuki et al., 1989). Also, palatalized articulation is the most frequently observed misarticulation among those children who achieved good velopharyngeal closure after the early repair of their cleft palate (Ainoda et al., 1985; Imai, 2003). Palatalized articulation, as defined by Okazaki (1982), is a backward distortion of the dentals or alveolars. It is characterized by articulatory contact of the dorsum instead of the tip or the blade with the posterior border of the hard palate (Okazaki et al., 1980, 1991; Michi et al., 1986). Okazaki (1982) and Okazaki et al. (1991) reported that palatalized articulation was seldom seen in children with soft cleft palate. Some reports described that the incidence of palatalized articulation in children with cleft palate only was significantly lower than in those having both cleft lip and palate (Ainoda, 1981; Yoshimasu et al., 1986; Suzuki et al., 1989). Although malformation of the palatal arch has been advocated as a possible cause of palatalized articulation (Okazaki et al., 1991), palatalized articulation is often noted even in children with a relatively good maxillary growth (Suzuki et al., 1989; Suzuki et al., 1994). Yamashita and Michi (1991) suggested that a habit caused by the cleft palate before surgical correction may be a factor in establishing an abnormally retracted tongue position during production of the dental and alveolar sounds.

It was hypothesized, therefore, that differences in tongue-use patterns associated with the abnormal preoperative structure may result in palatalized articulation. Furthermore, the OP might be expected to facilitate phonetic development in the preoperative oral cavity of an infant with cleft lip and palate. The purpose of this study was to investigate the effect of the OP from the perspective of the postoperative articulatory function.

The present study consisted of two parts. In the first part, the mobility of the tongue of infants with cleft lip and palate wearing the OP was evaluated by ultrasonography (USG) during sucking. In the second part, a long-term follow-up was conducted to evaluate postoperative articulatory function in those children with and without preoperative application of the OP.

TABLE 1 Characteristics of Subjects and Findings by USG

Group	*Case*	*Cleft Type*	*Sex*	*Age (mo)*	*Alveolar Cleft Width (mm)*	*L:H*
Normal infants	A	—	M	7	—	0.89
	B	—	F	7	—	0.90
	C	—	F	4	—	0.85
Infants with cleft lip and palate and without OP	D	UCLP	M	5	15.0	0.66
	E	UCLP	M	7	unknown	0.61
	F	UCLP	M	6	10.0	0.68
Infants with cleft lip and palate and OP	G	BCLP	F	6	3.2	0.73
	H	UCLP	F	6	10.5	0.59
	I	UCLP	M	5	12.6	0.54

Observation of Preoperative Tongue Movements During Sucking

Methods

Subjects

Six infants 5 to 7 months of age with complete cleft lip and palate served as subjects. Three of them, two with unilateral cleft lip and palate (UCLP) and one with bilateral cleft lip and palate (BCLP), used the OP. The three remaining subjects with UCLP did not use the OP. In addition, three normal infants served as a control group (Table 1). The birth weights of all infants were greater than 2500 g.

Procedures

This study followed the accepted ethical standards formulated in the Declaration of Helsinki. Informed consent was obtained from a parent of each subject before data collection.

Tongue movements of the subjects during sucking were observed by USG (U-sonic RT4600, Yokogawa Medical Systems, Tokyo, Japan). Smith et al. (1985) and Weber et al. (1986) first reported the ultrasonographic study of infants' sucking. Because ultrasound has no known adverse biological effect, infants and children can be safely examined. Furthermore, USG allows for real-time imaging of sucking in a natural position during feeding for the mother and infant without the use of contrast material (Bu'Lock et al., 1990; Sonies, 1991). In the present study, each mother held her infant in her arms as usual, and an ultrasonic probe was placed under the infant's chin. The mother then began to give the infant milk with a feeding bottle and favorite nipple. About 2 to 3 seconds after the onset of feeding, when the infant seemed to be constantly sucking, midsagittal B-mode images were recorded. Because previous reports (Bosma et al., 1990; Horikawa et al., 1995) and our preliminary observations in normal infants indicated that the tongue usually showed a cyclic movement of

0.7 to 1 second each, continuous images of at least 2-second duration (4 seconds on average) were recorded on a videotape recorder (SVO-5800, Sony Corp., Tokyo, Japan).

The images were then digitized at 30 frames per second. Each frame was printed with a laser printer, and the first author (K.S.) traced the contour of the surface of the tongue under the guidance of an engineer who had 20 years of experience with USG. An engineer who had 8 years of experience with USG judged the adequacy of the tracings. The tracings were compared and analyzed in temporal order. Frame-by-frame observations of the video images were also referenced to the tracings.

Results

In each printed image, the contour of the surface of the tongue and the contour of the feeding-bottle nipple were clearly identified, and the surface of the palate could be identified as the milk spread into the oral cavity.

In the normal controls, continuous rhythmical movements resembling peristalsis were observed on the surface of the tongue. Figure 2 shows one cycle of continuous movements of the tongue from a normal infant (case C). The front part of the tongue is displayed on the left of each print, and the line traces the surface of the tongue. The surface of the tongue started to continuously elevate from the front (frame 2) to the back to push the nipple. The rear portion of the tongue then descended and milk spread into the oral cavity. The tongue then remained in a flat position for a moment (frame 22–24) and then the front portion started to elevate again, beginning the next cycle.

The analysis was conducted in the same manner for the infants with cleft palate. Figures 3 to 5 are sagittal views of the tongues when milk was ejected from the nipples. In the cleft palate groups (Figs. 4 and 5), the extent of downward displacement of the rear portion of the tongue was greater than that in the normal infants (Fig. 3), regardless of the presence (Fig. 5) or absence (Fig. 4) of the OP.

For a statistical analysis, the distance from the probe to both the lowest point of the tongue (L in Fig. 3) and the highest point of the tongue (H in Fig. 3) were measured in these figures, and the ratio of L:H was calculated (Table 1). The first and second authors (K.S. and Y.Y.) measured the distances, and the correlation coefficient of 18 values measured by two raters was 0.99. The mean ratios of L:H in the two groups with cleft lip and palate (0.65 ± 0.04 for those without the OP and 0.62 ± 0.10 for those with the OP) were lower than that in the normal infants (0.88 ± 0.03), though the Kruskal-Wallis test revealed that the differences in the L:H ratio among the three groups did not reach a significant level ($H = 5.60$, $p = .06$).

It was also observed that all three subjects without the OP showed irregular peristaltic movements in which a part of the tongue showed simple repetitive movements so that the peristalsis was aborted or the direction of the tongue dorsum elevation appeared to be irregular. In contrast, none of the subjects with the OP showed such irregularity.

Evaluation of Postoperative Articulatory Function

Methods

Subjects

In this part of the study, postoperative articulatory function was evaluated in 17 children with complete cleft lip and palate (12 boys and 5 girls) for whom the OP was used at our hospital during the period from 1994 to 1999 (Table 2). The age of the participants at the time of the speech evaluation ranged from 4 years 1 month to 6 years 6 months. Six of the 17 children had bilateral clefts, and the remaining 11 children had unilateral clefts. All children had neither apparent mental retardation nor hearing impairment. Hearing was checked by visual reinforcement audiometry once before age 1 and then rechecked every 6 months. The OP was applied by 42 days after birth at the latest (18.6 days on average). OP application was continued until the time of palatoplasty in 7 children, whereas its use was discontinued between cheiloplasty and palatoplasty in 5 children (3½ months before palatoplasty on average) and at the time of cheiloplasty in 5 children (11.8 months before palatoplasty on average). Palatoplasty was performed between the ages of 1 year and 1 year 7 months (1 year 5 months on average). The surgical methods used were the modified Furlow method in 12 children and the mucosal grafts and flaps method in 5 children.

The control group consisted of 91 children with cleft lip and palate who were born during the period from 1981 to 1993 and underwent palatoplasty at the age of 1 year 5 months with no preoperative use of the OP (Table 3).

Evaluation of Articulatory Function

In all subjects, speech therapists assessed speech development every 3 to 6 months after birth. The last evaluation of articulatory function in the present study was made at 5 years 4 months of age on average or an average of 3 years 11 months after the palatoplasty (Table 2). ‘‘Last’’ means that the evaluation was made when normal adult articulation was observed in running speech in those children, showing no abnormal articulation. For those children with abnormal articulation, the term means that the evaluation was made immediately before any substantial speech training or secondary operations such as a fistulectomy or pharyngeal flap surgery. Because a longer observation was needed to identify a subject as a normal speaker, whereas less time was needed to judge if a subject needed speech therapy or a secondary operation, the average age at evaluation of these children with normal articulation was slightly higher than that of those with abnormal articulation.

For evaluation of the velopharyngeal as well as articulatory function, a test battery proposed by the committee on cleft

FIGURE 2 Traced surface of the tongue during normal sucking.

palate speech, the Japan Society of Logopedics and Phoniatics (Kumai et al., 1984; Ohira et al., 1993), was used. This battery consisted of speech evaluation (25 words, single syllables, and some sentences) and the use of a checklist of the articulatory organs. One speech therapist assessed articulatory function in each case on an outpatient basis, and the audio-recorded speech samples were further evaluated by three speech therapists, including the first author of the present paper (K.S.). Each therapist independently listened to and transcribed all samples by using the International Phonetic Alphabet. A rate of agreement among the listeners was obtained as a ratio between the number of agreed syllables and the entire syllables included in a 25-word test. The average rate of agreement among the three listeners was 87% for all syllables and 78% for palatalized articulation. The final judgment of the existence and the type of an abnormal articulation was made when at least two of the three judges agreed in their evaluation.

Results

Six of the seven subjects who used the OP continuously until the time of palatoplasty obtained normal articulation, whereas the remaining subject, who needed surgical closure of a residual fistula, developed glottal stops. Three of the other five subjects, in whom the use of the OP was discontinued sometime between the cheiloplasty and the palatoplasty, developed palatalized articulation. Among the remaining five subjects, in whom the use of the OP was discontinued at the time of cheiloplasty, two developed palatalized articulation and

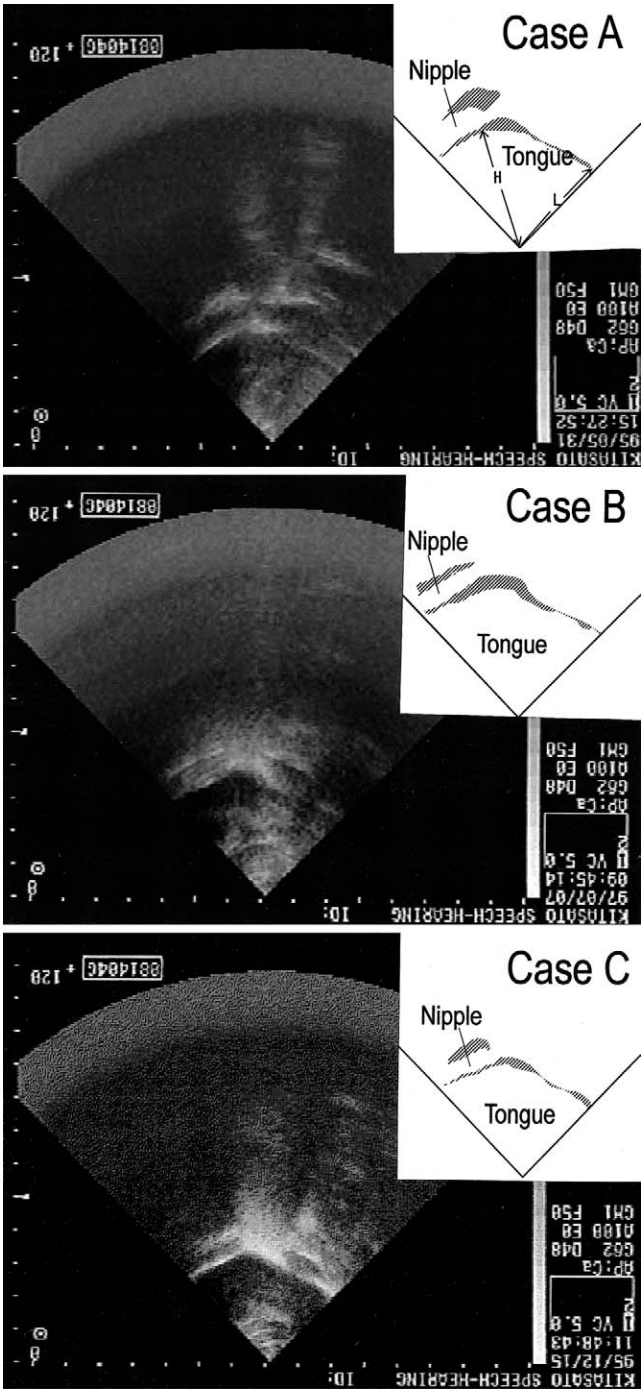

FIGURE 3 Sagittal view of tongue showing downward displacement of the rear portion at the time when milk is ejected from the nipple. Normal infants: cases A, B, and C.

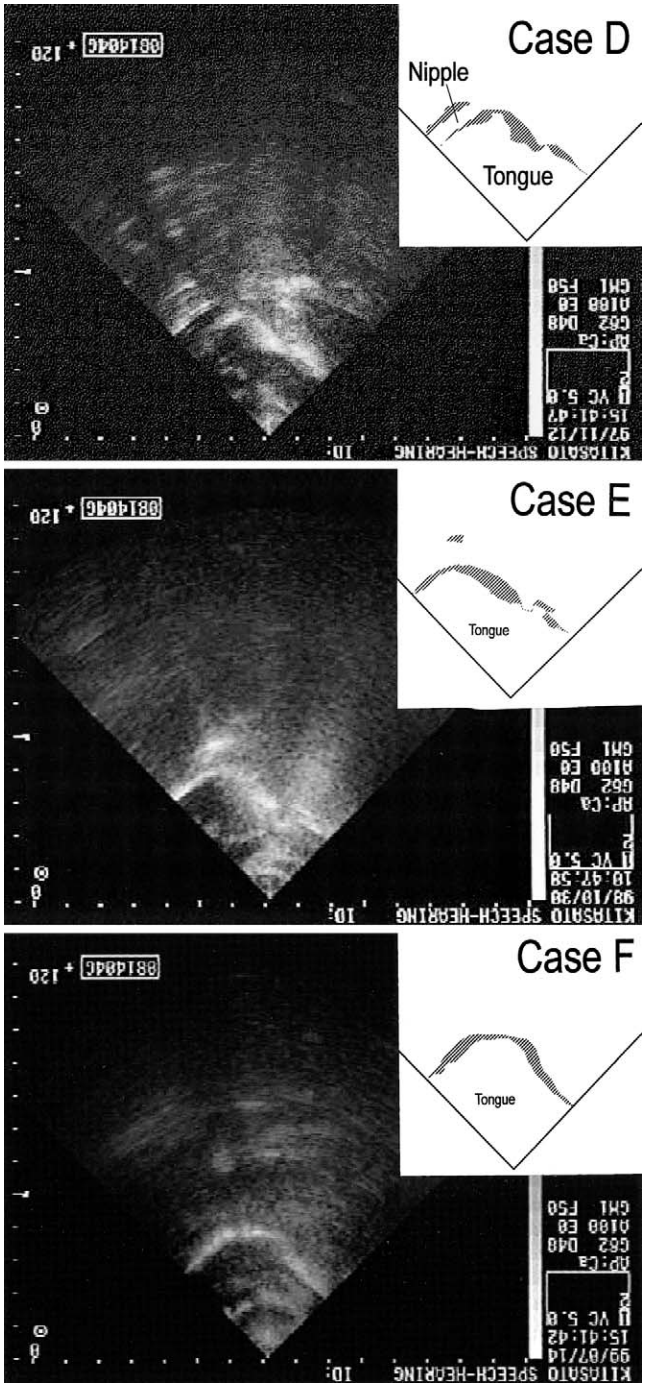

FIGURE 4 Sagittal view of tongue showing downward displacement of the rear portion at the time when milk is ejected from the nipple. Infants with cleft lip and palate and without OP: cases D, E, and F.

one, who needed both fistulectomy and pharyngeal flap surgery, developed glottal stops (Table 2).

All six subjects who continuously used the OP and needed no secondary operations (i.e., who displayed velopharyngeal adequacy and no oronasal fistula after the palatoplasty) obtained excellent articulation without speech therapy. Five of the nine subjects who discontinued use of the OP sometime before the palatoplasty and needed no secondary operations developed palatalized articulation and other misarticulations. The percentage of subjects obtaining excellent articulation was significantly higher (Fisher exact probability test, $p = .042$) in the group who continuously used the OP than in the group who discontinued use. However, there was no significant difference in the percentages of the subjects with disordered articulation between the group who discontinued the OP and the control group, who never used it (Table 3).

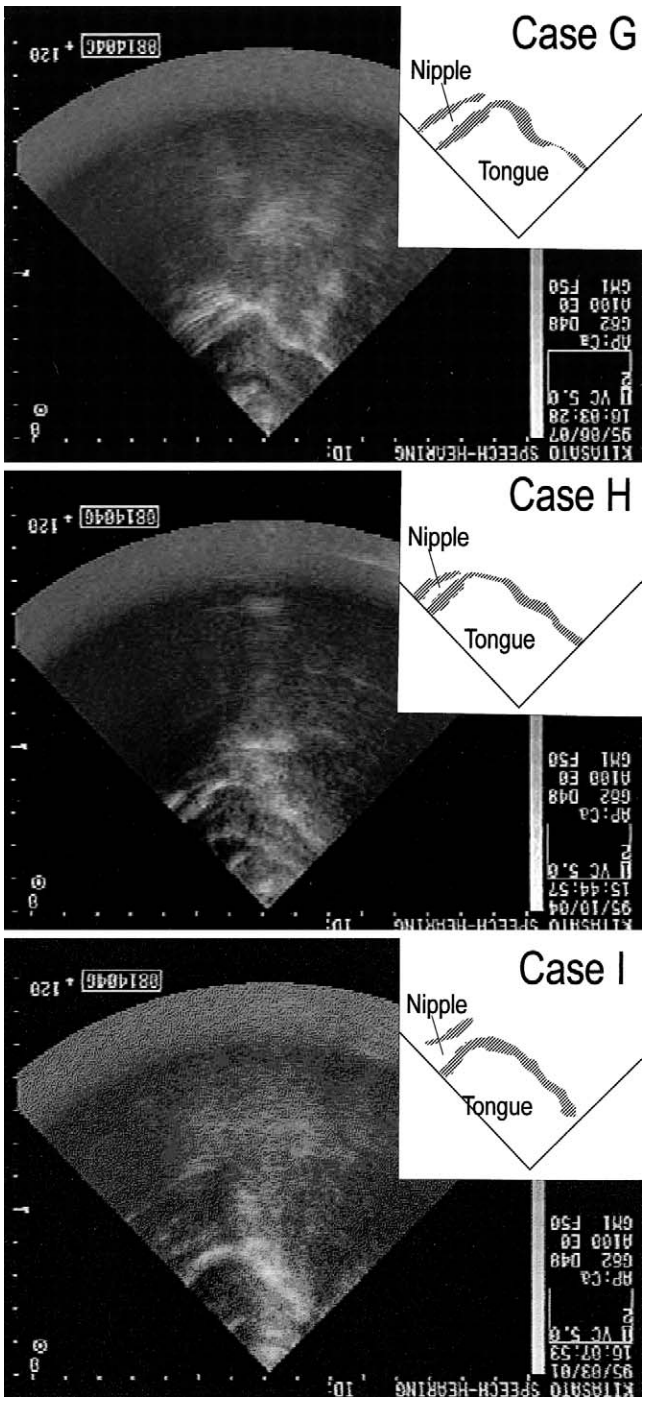

FIGURE 5 Sagittal view of tongue showing downward displacement of the rear portion at the time when milk is ejected from the nipple. Infants with cleft lip and palate and OP: cases G, H, and I.

DISCUSSION

A normal infant sucks milk by tightly closing the oral cavity at the tip of the tongue, thus sealing the base of the nipple anteriorly, and attaching the base of the tongue to the soft palate posteriorly, thus creating a negative pressure in the cavity (Ardran et al., 1958a, 1958b; Bosma et al., 1990; Eishima, 1991; Horikawa et al., 1995; Haishima et al., 1997). Infants

TABLE 2 Characteristics of Subjects Who Used OPs and Their Articulation

*Subject Group**	*No.*	*Sex*	*Cleft Type*	*Age at OP Start*	*End OP to Palatoplasty*	*Age at Palatoplasty*	*Palatoplasty Method*	*Age at Evaluation*	*Articulation*
A	1	F	UCLP	15 d	0 mo	1 y 6 mo	modified Furlow method	5 y 4 mo	excellent
	2	M	UCLP	30 d	0 mo	1 y 5 mo	modified Furlow method	6 y 4 mo	excellent
	3	M	BCLP	14 d	0 mo	1 y 3 mo	modified Furlow method	5 y 11 mo	excellent
	4	M	UCLP	23 d	0 mo	1 y 4 mo	modified Furlow method	6 y 5 mo	excellent
	5	M	UCLP	27 d	0 mo	1 y 6 mo	mucosal grafts and flaps method	5 y 7 mo	excellent
	6	M	BCLP	22 d	0 mo	1 y 4 mo	mucosal grafts and flaps method	6 y 6 mo	excellent
	†7	M	BCLP	27 d	0 mo	1 y 7 mo	mucosal grafts and flaps method	4 y 4 mo	glottal stops
B	8	F	UCLP	15 d	1 mo 24 d	1 y 5 mo	modified Furlow method	5 y 11 mo	excellent
	9	M	BCLP	10 d	5 mo 4 d	1 y 3 mo	modified Furlow method	5 y 10 mo	excellent
	10	F	BCLP	16 d	2 mo 19 d	1 y 7 mo	modified Furlow method	5 y 1 mo	palatalized articulation
	11	F	UCLP	20 d	2 mo 27 d	1 y 6 mo	modified Furlow method	4 y 9 mo	palatalized articulation
	12	F	UCLP	19 d	5 mo 14 d	1 y 7 mo	modified Furlow method	6 y 0 mo	palatalized articulation
C	13	M	UCLP	3 d	9 mo 7 d	1 y 0 mo	modified Furlow method	5 y 0 mo	excellent
	14	M	UCLP	7 d	16 mo 3 d	1 y 6 mo	modified Furlow method	4 y 7 mo	excellent
	15	M	UCLP	6 d	8 mo 10 d	1 y 0 mo	modified Furlow method	4 y 7 mo	palatalized articuation, substitution
	16	M	UCLP	42 d	12 mo 27 d	1 y 4 mo	mucosal grafts and flaps method	4 y 1 mo	palatalized articulation, lateral articulation
	†17	M	BCLP	20 d	12 mo 25 d	1 y 5 mo	mucosal grafts and flaps method	4 y 5 mo	glottal stops
Average				18.6 ± 9.7 d		16.6 ± 2.2 mo		64.0 ± 9.5 mo	

* A = subjects who used OP until palatoplasty; B = subjects who stopped OP between cheiloplasty and palatoplasty; C = subjects who used OP until cheiloplasty.
† The secondary operation was done after the evaluation.

TABLE 3 Characteristics of the Control Subjects and Their Articulation

Subjects, n	91
Sex, n	
Male	56
Female	35
Cleft type, n	
BCLP	21
UCLP	70
Age at palatoplasty, mo	17.3 ± 2.2
Method of palatoplasty	
Modified Furlow	8
Mucosal grafts and flaps	83
Age at evaluation, mo	60.0 ± 11.7
Articulation, n (%)	
Excellent	47 (51.6)
Abnormal	44 (48.4)
Palatalized articulation	27 (61.4)
Glottal stops	10 (22.7)
Lateral articulation	8 (18.2)
Other	11 (25.0)

with cleft palate have difficulty creating a negative pressure in the oral cavity because of a connection between oral and nasal cavities. In a study involving infants with cleft palate, Suzuki et al. (1997) used USG to observe an apparent large downward excursion of the rear portion of the tongue during sucking. They suggested that such an excursion compensated for the insufficient negative pressure.

In the present study, a greater downward excursion of the rear portion of the tongue during sucking was observed in infants with cleft palate regardless of the use or nonuse of the OP. This result would indicate that the OP was not effective for the creation of negative pressure in the oral cavity during sucking. The assumption was supported by the fact that those infants who used the OP required a special feeding nipple. It thus seems reasonable to speculate that the OP might not facilitate tongue-tip mobility, which has been reported in normal sucking. However, it was observed that irregular movements of the tongue during sucking were apparently suppressed in infants who used the OP. This might suggest that the OP plays a role as the roof of the oral cavity. By covering the cleft, the OP might prevent the tongue or the nipple from inserting into the cleft and inhibit excessive movements of the tongue during sucking.

In the second part of the present study, postoperative articulatory results were examined. Considering the better growth of the upper jaw, our medical team has used two types of palatoplasty. Recently, we have adopted the modified Furlow method for most of the patients with cleft palate because of its methodological simplicity. Suzuki et al. (1994) has reported that there is no significant difference between the articulatory results for the two types of palatoplasty. Good jaw growth results have been reported (Torikai et al., 1997; Udagawa, 2001). Under these circumstances, the present study included all subjects with the two different surgical procedures. Although the number of subjects was relatively small, the contrast of articulatory results between those who continuously used the OP and the others who discontinued it appeared clear. The percentage of subjects with disturbed articulatory function in the latter group was comparable with that in the control group, who never used the OP. Articulatory function of the control group seems to represent the average of the Japanese children with cleft palate. The result of the present study suggests that preoperative use of the OP appeared to have some effects on preventing postoperative abnormal articulation if it were continuously worn until the time of palatoplasty.

Palatalized articulation, which is the most frequently observed misarticulation in Japanese children with cleft palate, is partially similar to the middorsum palatal stops reported by Trost (1981). The former may be a backed realization of the latter, including fricatives and affricates. Both use the dorsum instead of the tip or the blade of the tongue for articulation of dentals or alveolars. Furthermore, several reports have described that the dental or alveolar sounds are rare in the babbling or the early speech of children with cleft palate (O'Gara and Logemann, 1988; Estrem and Broen, 1989; Chapman and Hardin, 1992; Suzuki et al., 1993). Harding and Grunwell (1996) included the backing pattern in ''cleft-type developmental processes'' and indicated that alveolar targets are the most vulnerable. Lohmander-Agerskov et al. (1995) indicated that apicodental pressure consonants are retracted to a dorsovelar place of articulation in many children with a cleft in the hard palate. It is thus assumed that using the tip or blade of the tongue for articulation might be a common difficulty for these children, regardless of language. Konst et al. (2003) suggested that the improved opportunity in infants with the OP to practice alveolar consonants may relate to the improved phonological development between 2 and 3 years of age. In the first part of the present study, USG images suggested that the OP could inhibit irregular movements of the tongue during sucking, though it was not effective for creating a negative pressure. If continuous wearing of the OP would have some effect on preventing palatalized articulation, the OP might also have provided an infant with some appropriate conditions for facilitating normal movements of the tongue during the preoperative period.

Further study of more subjects should be expected to allow for a clearer conclusion of the effect of the OP on postoperative articulatory function.

Conclusion

Preoperative use of the OP was applied to infants with complete UCLP or BCLP who were scheduled for a one-stage palatoplasty. Ultrasonography images of tongue movements during sucking revealed that (1) the OP, which occluded the cleft only at the level of the hard palate, was not effective for the creation of a negative pressure in the oral cavity; and (2) the OP inhibited irregular movements of the tongue. Long-term follow-up of postoperative articulatory function revealed that continuous use of the OP appeared to be effective in preventing palatalized articulation. By covering the cleft, the OP might prevent the tongue or the nipple from getting into the

cleft irregularly and inhibit excessive movements of the dorsum, which may be a cause of palatalized articulation.

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