Unoperated Adult Cleft Lip and Palate: Changes in Form and Function after Operation

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This report describes the occlusion and craniofacial morphology—together with changes in tongue activity during swallowing before and after surgical correction—in a 26-year-old female patient with unilateral cleft lip and palate. Cross-bites were not observed before or after repair. Lateral cephalometric findings revealed a convex profile with a normal anterior cranial base length, a flat mandibular plane, and a short lower-anterior face height. Preoperative cineradiographic appraisal showed atypical tongue activity during swallowing. The dorsum of the tongue remained high to seal the cleft. The bolus was kept on the floor of the mouth and passed along the lateral margins of the tongue. The swallow duration, as determined by EMG of the tongue, was extremely long when compared with infant-operated cleft lip and palate patients and to noncleft subjects. Postoperatively, tongue function was similar to that in both cleft patients in whom closure was performed in infancy and to noncleft controls.

KEY WORDS: *cleft lip, cleft palate, cephalometry, cineradiography, tongue, swallowing, EMG.*

A recent case of unoperated adult unilateral cleft lip and palate provided interesting findings relating to form and function of the orofacial area. This report describes the occlusion, craniofacial morphology, and changes in tongue activity during swallowing before and after cheiloplasty and palatoplasty in an adult female patient.

CASE PRESENTATION

The patient was the fourth of five children, and a family history of cleft lip and palate was not identified. Although she had the opportunity to attend hospital cleft palate clinics, she did not receive surgical or dental care until she was 26 years old, and she had never worn an obturator (Fig. 1). Her height and weight (157 cm and 59 kg, respectively) were within normal ranges for Japanese adult females.

When the patient was 27 years and 3 months old, the cleft lip was repaired by a combined Millard-Cronin method. One year later, at 28 years and 3 months, surgical closure of the cleft palate was performed along with a superior-based pharyngeal flap operation (Fig. 2).

Records consisting of intraoral and facial photographs, dental casts, lateral cephalograms, and cineradiographs, as well as EMG studies of tongue activity during swallowing, were obtained before cheiloplasty and at eight and 18 months after palatoplasty.

Dental Cast Analysis

Preoperatively, crossbites were not observed in the anterior or lateral segments (Fig. 3). After repair, no changes were observed in the length or width of the upper dental arch. Changes in dental arch, which form postoperatively, usu-

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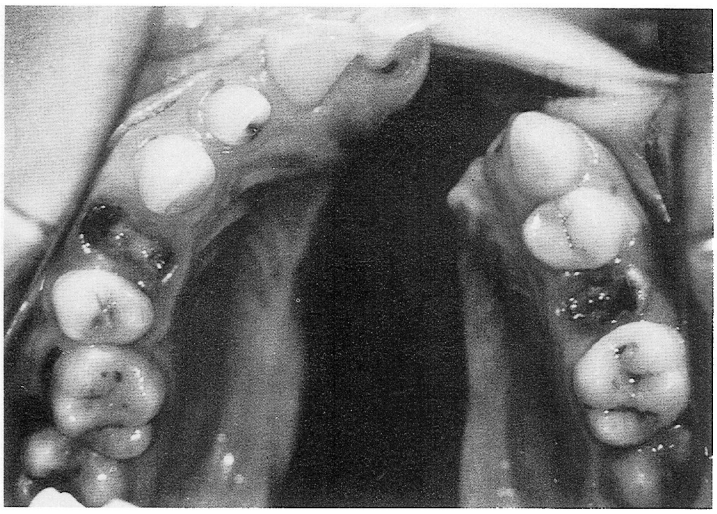

FIGURE 1 Palatal view of the unoperated cleft palate in a 26 year old patient.

ally in the infant surgical patient, did not occur, except for some minor spontaneous alignments of the maxillary central incisors (see Fig. 3). The patient demonstrated a Class II canine relationship. Molar relationships could not be determined because the lower molars had been lost, owing to decay.

Cephalometric Analysis

The preoperative lateral cephalogram revealed a convex profile with a normal length of the anterior cranial base. The maxillary depth—the dimension between the anterior and posterior nasal spines—was longer than the mean obtained for control subjects but was within a normal range. The maxilla was protruded relative to the anterior cranial base; this suggests a skeletal Class II relationship (Fig. 4). An appropriate Wits appraisal (Jacobson, 1976) could not be made because the occlusal plane was altered by the loss of the lower molars. A short maxillary height relative to the sella-nasion (S-N) plane was quantified. A flat mandibular plane and a short lower anterior face height were observed. A steep mandibular plane, one of the characteristics of unoperated cleft lip and palate patients, was not observed. The axial inclination of the maxillary and mandibular incisors—as indicated by the angle of the maxillary incisor to the Frankfort horizontal (FH) plane, the angle of the mandibular incisor to the mandibular plane, or the Frankfort-mandibular incisor angle (FMIA)—was within normal limits. Postoperatively, quantifiable changes were not observed in these cephalometric measurements.

Cineradiographic Analysis

To evaluate swallowing patterns without an obturator and to observe how the tongue

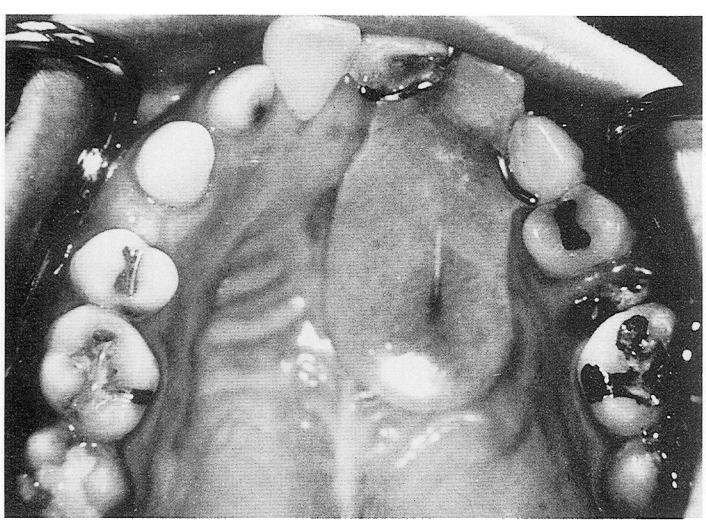

FIGURE 2 Postoperative palatal view of a 29 year old patient. An obturator was placed to close a fistula.

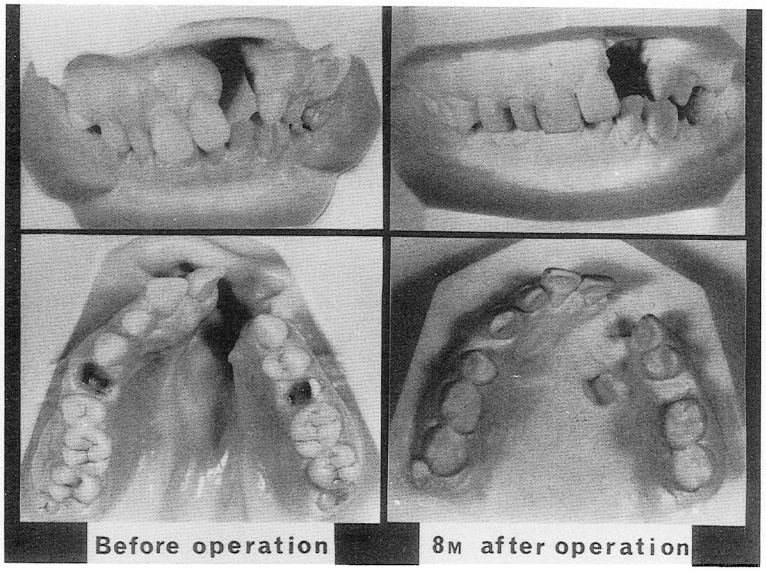

FIGURE 3 The occlusion and upper dental arch form, shown before and 8 months after operation. Neither crossbites nor collapse of the dental arch was observed postoperatively.

adapted to the new oral environment after palatoplasty, a series of swallows was recorded cineradiographically. Thin lead markers were pasted to the tip of the tongue and 40 mm behind the tip on the dorsum. A small amount (4 to 5 ml) of orange juice containing barium sulfate was used as a test food. The patient was seated on a chair, and 35-mm film cineradiographs were exposed at 24 frames/sec in the lateral and anteroposterior planes.

Before surgical repair, a cineradiographic appraisal revealed atypical tongue activity during swallowing. The dorsum was elevated to seal the cleft, and the bolus was maintained on the floor of the mouth during the oral phase of the swallow. The bolus passed along the lateral margins of the tongue. The phenomenon of "free fall" described by Shelton et al (1966) was observed at the end of the oral phase. This series of tongue movements during swallowing was different from the typical swallowing pattern obtained for a noncleft subject (Figs. 5 and 6) and from that of 250 normal young adults as reported by Ardran and Kemp (1955).

After lip and palatal repair, the bolus was found on the floor of the mouth at the initiation of swallow. However, 8 months after palatoplasty, some of the bolus was located on the dorsum of the tongue during the oral phase of swallowing (Figs. 7 and 8). Tongue exercises were not utilized. Free fall was still observed at this stage, although a "stripping action" of the tongue against the soft palate and pharyngeal area was also seen. Eighteen months after palatoplasty, cineradiographic studies revealed a near-normal swallow pattern. Although the bolus was pooled on the floor of the mouth at the initiation of swallow, most of it was brought to a depression on the dorsum during the oral phase; a stripping action of the tongue was observed during swallowing, and the free fall phenomenon was diminished.

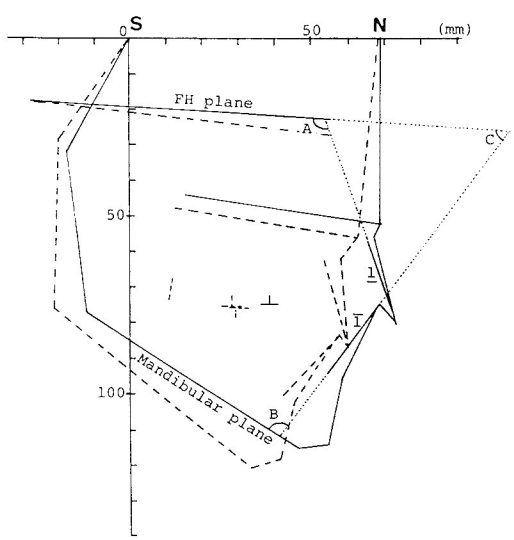

FIGURE 4 Profilograms superimposed on the S-N plane registered at S. $\underline{1}$, axis of the maxillary incisor; $\overline{1}$, axis of the mandibular incisor; A, angle of the maxillary incisor to the Frankfort horizontal (FH) plane; B, angle of the mandibular incisor to the mandibular plane; C, Frankfort-mandibular incisor angle (FMIA). Solid line = before operation; dashed line = mean of noncleft subjects (N = 20).

Tongue EMG Analysis

To identify the duration of the swallow, tongue EMG was recorded simultaneously with cineradiographic examination, which generated time pulses. Miniature surface electrodes were applied to the dorsum of the anterior portion of the tongue. This site was selected because of the

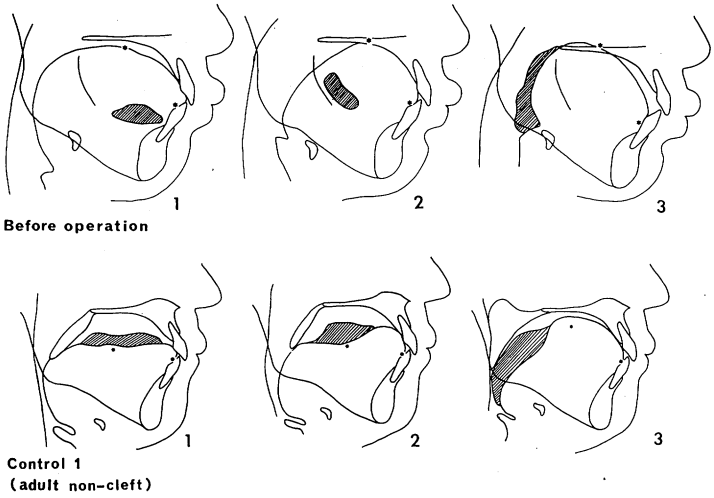

FIGURE 5 Cineradiographic appraisal of the lingual activity during swallowing. Lateral cineradiographic frames indicate an atypical swallow pattern before operation and a normal swallow pattern of a noncleft adult (Control 1). Note the differences in bolus position (shaded area) and in tongue configuration. The asterisks indicate lead markers at the tip of the tongue and 40 mm behind the tip.

ease of placement of the surface electrodes. The area is of particular interest when adaptation of the tongue to the altered environment is studied (Subtelny, 1970). The electrodes consist of highly-conductive silver paste, small rubber caps, and platinum fine wires (Fig. 9). With this method, the EMG activity of the tongue during various functions can be monitored without causing anxiety in or discomfort to the patient. The electrodes showed excellent stability, and their performance was consistent and reproducible in later trials (Yoshida et al, 1982).

Electromyographic activity recorded in this way was generated from the intrinsic tongue musculature (Yoshida, 1986). When EMG activities of intrinsic and extrinsic muscles of the tongue during swallowing were simultaneously obtained, intrinsic-muscle activity lasted slightly longer than that of the extrinsic muscles. Since intrinsic muscle movements alter the shape of the tongue, longer activity suggests that changes in tongue shape during swallowing start before that of extrinsic muscles and that the tongue returns to its original shape after extrinsic muscle activity has ended.

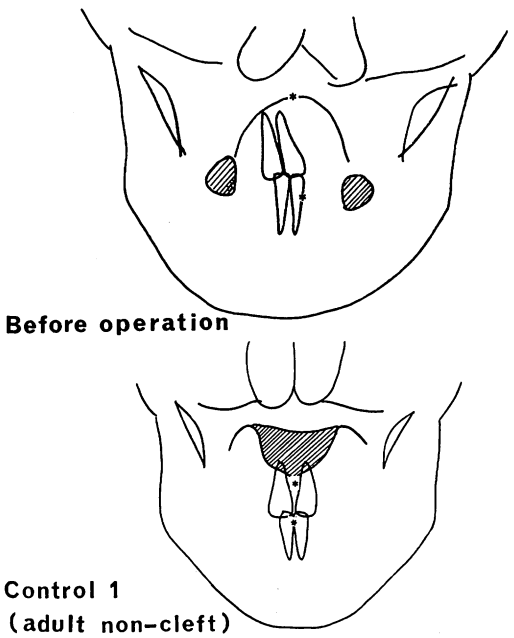

FIGURE 6 Anteroposterior cineradiographic frames indicate an atypical swallow pattern (top) and a normal swallow pattern (bottom). By simultaneous recording techniques, the frames coincide with Phase 2 of the lateral cineradiographic records.

Onset and completion of the swallow were determined by measuring the duration in the mean EMG curve, which was evaluated by a computer-assisted system. During swallowing, the mean EMG curve showed a high activity period at the beginning, which may be referred to as the swallow phase (*D-1* in Fig. 10), followed by a low activity period, which may be called the postswallow phase. In our previous study, great variation was noted in the interval of significant electric potentials after the swallow/postswallow phase (*D-2* in Fig. 10) among 10 subjects with normal occlusion and among independent swallows of a single subject (Takada et al, 1985). According to Yoshida (1986), the standard deviation (SD) of the swallow duration (*D-1*), as determined by the same method shown in Fig. 10, which is described later, was less than that of swallow/postswallow duration (*D-2*). One SD of D-1 duration for five swallows in 22 subjects with normal occlusion was only 120 msec, whereas the SD for D-2 duration was 832 msec.

For this reason, swallow duration (*D-1* in Fig. 10) was measured in this report. The onset and end of the swallow were determined by the following method (Fig. 10): EMG of the tongue at rest was sampled 20 times every 10 msec for

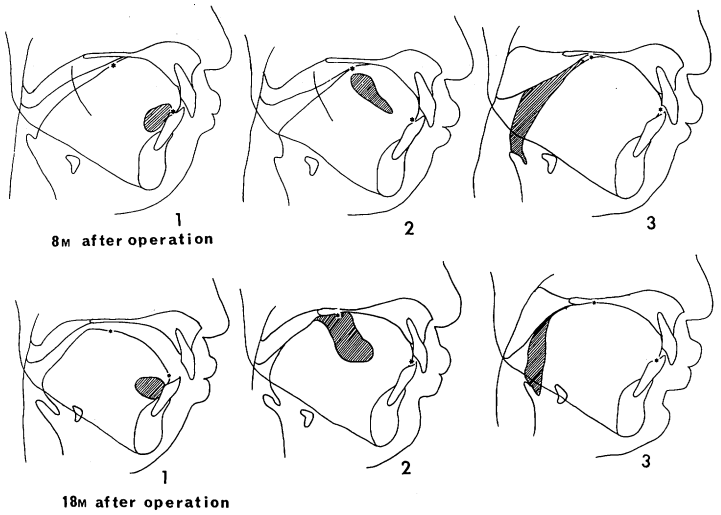

FIGURE 7 Cineradiographic appraisal of the lingual activity during swallowing 8 and 18 months after operation. Lateral cineradiographic frames indicate an improvement of the swallowing pattern after the surgical repair of the cleft lip and palate. The bolus was placed and taken from the floor of the mouth on the dorsum of the tongue during Phase 2.

200 msec (α), and the mean (Mα) and SD for EMG voltage across the 20 samples were computed. The onset of lingual activity for a swallow was identified when the EMG activity exceeded Mα plus 2 SD and when that level of activity was maintained for at least 500 msec.

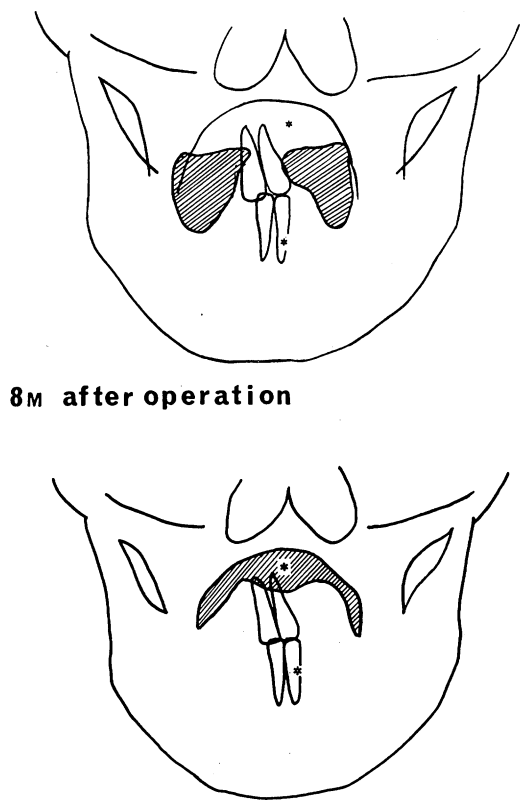

FIGURE 8 Anteroposterior cineradiographic frames that coincide with Phase 2 of Figure 7, showing that the bolus was placed on the dorsum of the tongue.

The postswallow phase was considered to have ended when EMG activity fell below a value 2 SD above the resting mean (Mα) and remained there for at least 500 msec. From this point in the reverse time direction, mean (Mβ) and SD of the EMG activity for the last period of the postswallow phase were calculated from 20 sampled points every 10 msec for a total of 200 msec (β). When the EMG activity became larger than Mβ plus 2 SD for at least 500 msec in the reverse time direction, the end of the swallow phase was identified.

Before palatoplasty, the duration of a swallow for this patient, as determined by tongue EMG, was longer than the typical swallow of an adult noncleft subject (Fig. 11). According to Yoshida (1986), the mean duration of lingual activity (*D-1*) for five swallows in the same subjects described before was 2008 ± 120 msec. Mean and SD of the duration were based on several acts of swallowing for each subject (Fig. 11). A significant decrease in the duration of a swallow was noted postoperatively, and the swallow duration resembled that of adult noncleft subjects or adult unilateral cleft lip and palate (UCLP) patients who had palatoplasty in infancy (Fig. 11).

DISCUSSION

It has been suggested that the dentofacial characteristics of adult patients with unilateral cleft lip and palate do not differ significantly from noncleft subjects (Mestre et al, 1960; Ortiz-Monasterio et al, 1966; Bishara et al, 1985; Bishara et al, 1986). However, in the case described in this report, the maxillary complex was well developed in the anteroposterior direction, and the skeletal profile was convex. These findings were different from the concave profile

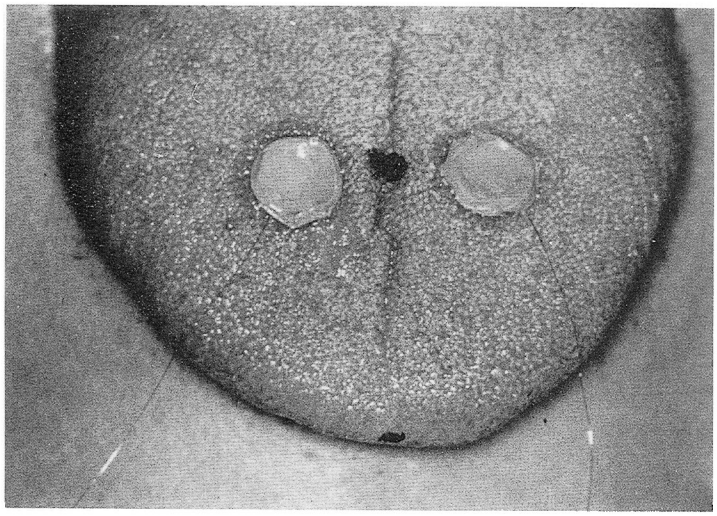

FIGURE 9 Tongue surface electrodes (Yoshida et al, 1982).

and underdeveloped maxilla that are often seen in patients who have received a one-stage surgical repair in infancy.

As to the vertical position of the maxilla, Mestre et al (1960) observed that unoperated cleft-palate subjects have normal vertical dimensions in both the posterior and anterior aspects of the maxilla. Dahl (1970) described 153 subjects with primary and secondary cleft palate. In 36 unoperated subjects, upper facial height was short, especially in the posterior region. Dahl (1970) suggested that the posterior height of the upper face was reduced after palatoplasty in subjects with combined clefts of the lip and palate. However, no logical explanation for this observation was provided. The present unoperated patient showed less vertical growth in the anteroposterior portions of the maxilla, when compared with noncleft subjects. This finding may indicate a reduced growth potential in the vertical direction of the maxillary complex. The low mandibular plane angle that was found may be related to the higher position of the maxillary complex and to the loss of the lower molars, owing to caries. A high mandibular plane angle or an obtuse gonial angle was reported, however, as a common feature of cleft-palate cases (Dahl, 1970; Chierici et al, 1973; Bishara et al, 1985; Bishara et al, 1986).

The cineradiographic and EMG evaluations of lingual activity during swallowing pertain to adaptation of the tongue to the altered morphology of the palate. The results support the concept that lingual function changes if its environ-

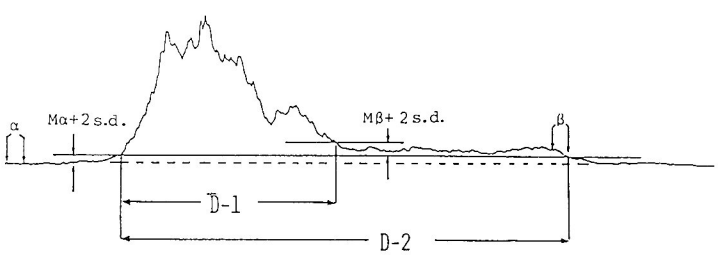

FIGURE 10 Method for the measurement of swallow duration in the mean EMG curve obtained from lingual activity by surface electrodes. Analysis was made by a computer-assisted system. α, Tongue EMG at rest was sampled 20 times every 10 msec for 200 msec, and the mean and standard deviation (SD) for EMG voltage across the 20 samples were computed; Mα, mean voltage at rest; β, tongue EMG at postswallow phase was sampled 20 times every 10 msec for a total of 200 msec from the point of its end in the reverse direction, as well as the mean and standard deviation (SD) were computed; Mβ, mean voltage of postswallow phase; D-1, swallow duration (see text); and D-2, swallow/postswallow duration (see text).

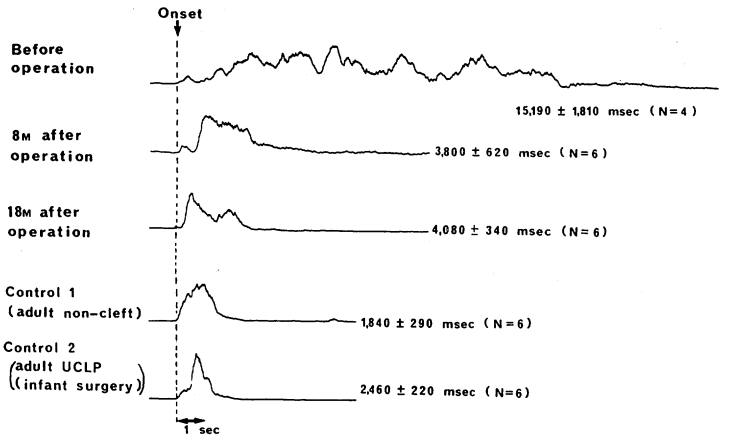

FIGURE 11 Duration of the lingual activity during swallowing as determined by a mean EMG curve. The top three EMGs were obtained from the patient. Note the extremely long swallow duration before palatoplasty compared with shorter durations, 8 and 18 months, after operation. Each of two EMGs shown in the lower part represents a typical swallow pattern from an adult noncleft subject and an infant-operated cleft lip and palate patient. N = number of swallows. The mean and SD for swallow durations are shown for each record.

ment is altered, as proposed by Subtelny (1970). Kawamura (1961) suggested that sensory modalities from the tongue, such as touch, play an important part in the feedback mechanisms that influence movement. Yokota et al (1974), on the basis of experimental studies in the cat, reported that tactile stimulation of the anterior part of the dorsum of the tongue had an effect on the extrinsic lingual musculature. These previous reports may provide evidence of a neurophysiologic basis for the changes in lingual motility observed after palatoplasty. Even in adult subjects, sensory information from the tongue is thought to elicit coordinated lingual movements. For speech production, compensatory movements of the tongue in an adult subject with unrepaired cleft palate were interpreted on the basis of oral motor skills (Fletcher, 1985). Higher center control is also considered to play an important role in the modulation of lingual activity (Lowe, 1981).

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