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# CONGENITAL ANKYLOGLOSSIA WITH DEVIATION OF THE EPIGLOTTIS AND LARYNX: SYMPTOMS AND RESPIRATORY FUNCTION IN ADULTS

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We operated on 38 adult patients with congenital ankyloglossia with deviation of the epiglottis and larynx. The results were as follows. 1) Most patients had Angle's class III malocclusion, irregular alignment of the upper teeth, and high hard palate. 2) Fifty percent of the patients in our study population had obstructive respiratory failure. Their vital capacity increased significantly after the operation, but changes of forced expiratory volume in 1 second were not prominent. 3) Subjective symptoms of this disease were stiffness of the shoulders, a cold feeling in the extremities, an obstructed feeling in the throat, insomnia, fatigue, dry skin, irritability and/or anxiety, and nervousness. These improved postoperatively. 4) Objective symptoms included snoring, muscle cramps, difficulty in playing wind instruments, hoarseness, and incorrect articulation. The objective symptoms, except for incorrect articulation, improved postoperatively.

KEY WORDS — ankyloglossia, insomnia, larynx, occlusion, respiration.

## INTRODUCTION

We have previously demonstrated that ankyloglossia is not only a forward dislocation of the tongue, but that it is also accompanied by an upper-forward dislocation of the epiglottis and larynx, and that newborn and suckling infants with this disease suffer from various degrees of dyspnea. Moreover, we reported on the symptoms and signs of congenital ankyloglossia in these children and, more important still, the relationship we think there is between this disease and sudden infant death syndrome.[1-6] In the past year we have treated several adult patients with ankyloglossia, and in this paper we report on the symptoms, signs, and state of respiratory function in adults with congenital ankyloglossia with deviation of the epiglottis and larynx (hereinafter called ankyloglossia).

## MATERIALS AND METHODS

Thirty-eight patients who visited the Otorhinolaryngology Division of the Mukai Clinic between September 1989 and October 1990 were judged to have ankyloglossia. Ankyloglossia was diagnosed by the criteria we previously reported.[2-6] All of them underwent correction of the tongue, epiglottis, and larynx.[7,8] These structures were moved down and backward by cutting the front bundles of the genioglossus muscles and the same level of the lingual septum. We studied the patients' occlusion, the state of their larynges, and their respiratory function before and after the operation. We recorded their chief complaints and their comments after the operation.

*Occlusion.* We used Angle's classification for malocclusion, that is, class I for neutroclusion, class II for distoclusion, and class III for mesioclusion.[9]

*Larynx.* We observed patients' larynges with laryngofiberscopes (Machida ENT 30 II, Yokohama, Japan) via the nose and classified them into 2 categories based on the extent of the vocal folds observed during phonation. In a type A larynx, more than 50% of the vocal folds was observable during phonation. In type B, less than 50% of the vocal folds was seen because they were obscured by the aryepiglottic folds and corniculate and cuneiform tubercles (Fig 1).

*Respiratory Function Tests.* We measured the patients' vital capacity (VC) and forced expiratory volume in 1 second (FEV1) with an autospirometer (Autospiro 100, Minato Medical Instruments Co, Tokyo, Japan). The VC measured with this spirometer can be expressed in liters or volume percent according to the subject's age, sex, and height. For data standardization we expressed VC in volume percent. According to the measurement obtained, the spirometer automatically classified each patient as having normal, obstructive, or restrictive respiratory function, or a combination of both obstructive and restrictive. We compared the preoperative and postoperative conditions of the respiratory function by means of the spirometer.

*Statistical Analysis.* For statistical analysis of the

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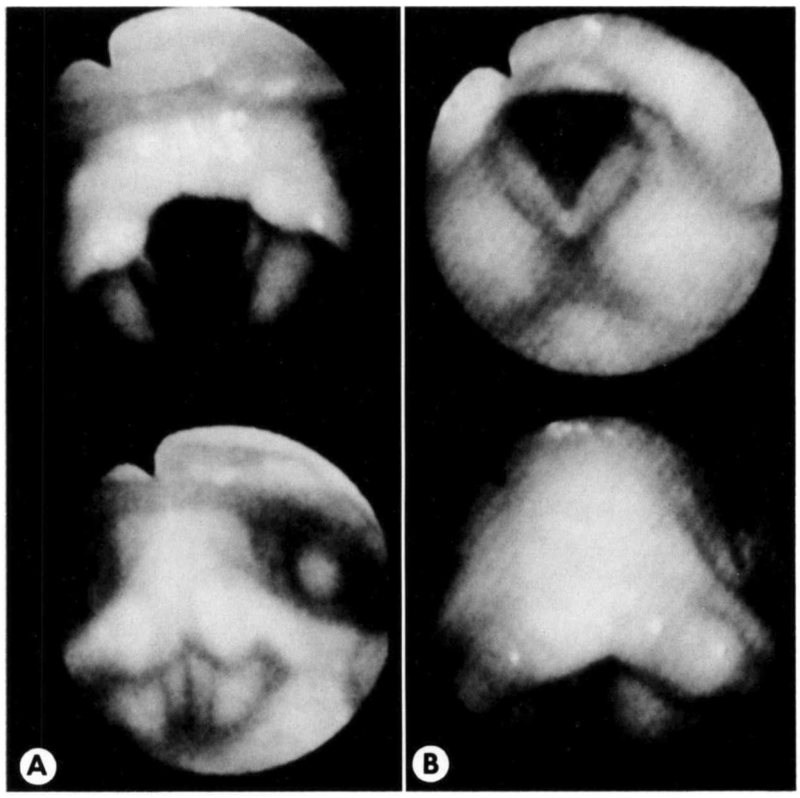


Fig 1. Laryngeal types. Top — during respiration, bottom — during phonation. A) Type A. B) Type B.

data we used McNemar's test, Student's paired *t* test, and the Stuart-Maxwell test.

## RESULTS

Thirty-eight patients, 18 male and 20 female, were examined for this study. The age range was from 13 to 58 years old, with an average of 30 years and an SD of 9 years. One patient had undergone frenotomy as an infant.

*Occlusion.* Six patients (16%) had class I malocclusions; of these, 2 patients had received orthodontic treatment. The other 32 patients (84%) had class III malocclusions. There were no class II malocclusions. Besides malocclusion, all the patients had incisor and canine teeth that were large in comparison with the other teeth. Their upper teeth were not aligned (Fig 2), and they also had a cupolalike, high hard palate.

*Larynx.* Before the operation, 13 patients (34.2%) had a type A larynx, and 25 (65.8%) had a type B. After the operation, 8 patients showed a modification of the vocal folds' appearance during phonation, and the numbers changed to 21 (55.3%) for type A and 17 (44.7%) for type B. The statistical analyses (McNemar's test) revealed that the surgery had significantly affected the state of the larynges ($p < .02$).

*Respiratory Function Test.* Before the operation the average VC of the patients was 93.2%, with an SD of 18.8%, and the average FEV1 was 60.6%, with an SD of 23.5%. The postoperative value for VC was 107.3%, with an SD of 18.4%, and that of FEV1 was 66.7%, with an SD of 21.1% (Fig 3). The changes in VC were statistically significant by paired *t* test ($p < .0001$). Postoperative diagnoses by the Autospiro 500 were as follows: normal, from 8 cases before surgery to 20 cases; obstructive, from 19 to 15; restrictive, from 5 to 2; and combined, from 6 to 1 (Fig 4). Statistical analysis (Stuart-Maxwell test) revealed that surgery was effective in cases with obstructive and/or restrictive respiratory failure ($p < .005$).

*Chief Complaints.* Patients presented with the following complaints. We divided them into subjective and objective chief complaints. Subjective complaints were as follows: stiffness of the shoulder (21 cases), an obstructive feeling in the throat (20 cases), a cold feeling in the extremities (11 cases), insomnia and/or the feeling of not sleeping well (5 cases), and fatigue (2 cases). Objective complaints were as follows: incorrect articulation (5 cases), muscle cramps (2 cases), snoring (2 cases), difficulty in tonguing while playing the flute (1 case), and hoarseness (1 case; this patient had laryngeal polyps). The subjective complaints improved postoperatively. Of the objective complaints, snoring, difficulty in tonguing, muscle cramps, and hoarseness improved postoperatively, while problems with articulation were not affected.

*Patients' Comments After Operation.* The postoperative comments varied widely. Twenty of 38 pa-

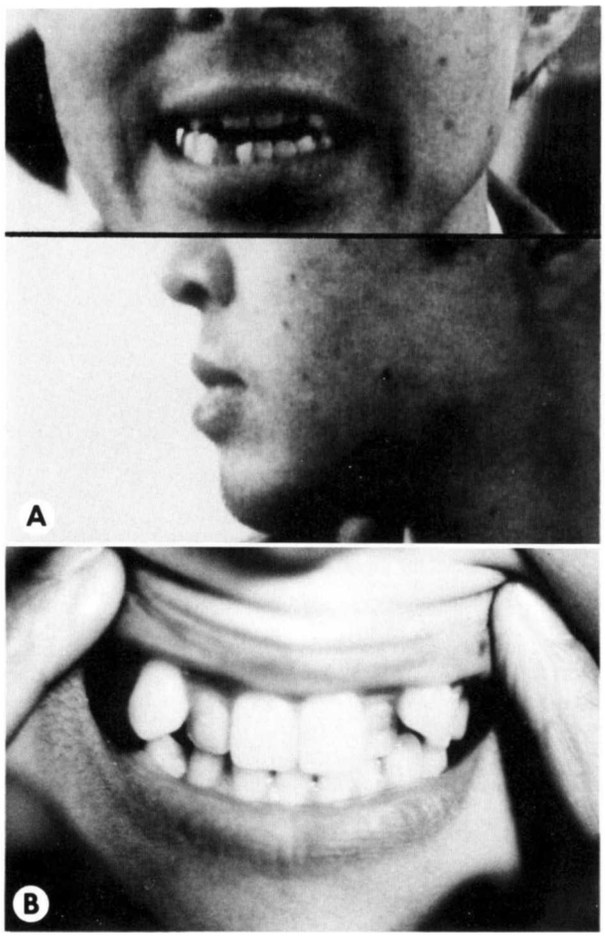


Fig 2. A) Observed Angle's class III mesioclusion. B) Malaligned upper teeth with large maxillary central incisors and canine teeth.

tients said they could breathe more easily. They could phonate better and for a longer time. Some felt their voices had become clearer (16 cases). Some could sleep better and/or felt rested in the morning (16 cases). Some felt their skin had turned brighter and healthier (11 cases). Some felt their throats to be more open (12 cases). Some had become calmer (2 cases). One patient had lost her feeling of anxiety and nervousness. According to their parents' observations, 2 patients who, before being operated on, showed signs of irritability did not have this sign anymore.

## DISCUSSION

*Occlusion*. Eighty-four percent of the patients had Angle's class III malocclusion. Two patients out of 6 with class I malocclusion had orthodontic treatment. In addition, they had irregular tooth alignment, with large incisor and canine teeth and a cupolalike hard palate. On the basis of these observations we can say that ankyloglossia, along with an Angle's class III malocclusion, brings about irregular tooth alignment and the development of a high palate. In a report of a patient with ankyloglossia, Takasu[10] noted that the patient had irregular alignment of the upper teeth and especially of the canine teeth. Tuerk and Lubit,[11] on the other hand, reported that patients with ankyloglossia develop an infantile swallowing habit and have an open bite and Angle's class III malocclusion. They warned that the ankyloglossia greatly affects the shape and growth of the skull. We observed that some patients who could not bite with their molar teeth could do so after the operation. We also saw a patient who could not bite *sembei* (hard rice cakes) until she was operated on. We think this is probably because ankyloglossia interferes with the movements of the jaw. Besides the patient who said that she could not bend her back without discomfort, other patients also said that their back pain had disappeared after the operation; this leads us to think that ankyloglossia not only causes malocclusion but also brings about an imbalance of the spine. High hard palates were observed, also. We have already reported that infants with ankyloglossia have a high hard palate, and apparently it continues throughout life.

*Larynx*. According to our classification of the larynx, the A:B ratio before surgery was 13:25, and after the operation it was 21:17. This reveals that adults with ankyloglossia have the same deviation of the larynx as we previously observed in infants. After the operation infants could cry louder, and among the 38 patients reported here, 17 said their voices had become louder and clearer. These improvements of the voice were observed in those patients who went from type B to type A, as well as in those with type A who experienced further improvements. There was 1 patient with laryngeal polyps among the reported cases. We have seen 2 other patients with ankyloglossia who had laryngeal polyps (not included in these data). All of them underwent surgery to correct ankyloglossia, the epiglottis, and the larynx. Three months after surgery the polyps on the vocal folds had disappeared. Probably, when patients with ankyloglossia phonate, the vocal folds in the deviated larynx receive an excessive stress load and easily develop polyps.

*Respiratory Function Test*. The most prominent change in the respiratory function was an increase of the VC of the lung, an average of 14.1% that was statistically significant. The results were so amazing that at the beginning we could not believe what we were seeing. As we have already reported[1-3] in newborn and suckling infants, we observed that as soon as the operation finished the patients' skin turned pink and bright, and their features turned more vivid. We thought that these changes were due to the disappearance of the respiratory failure caused by the

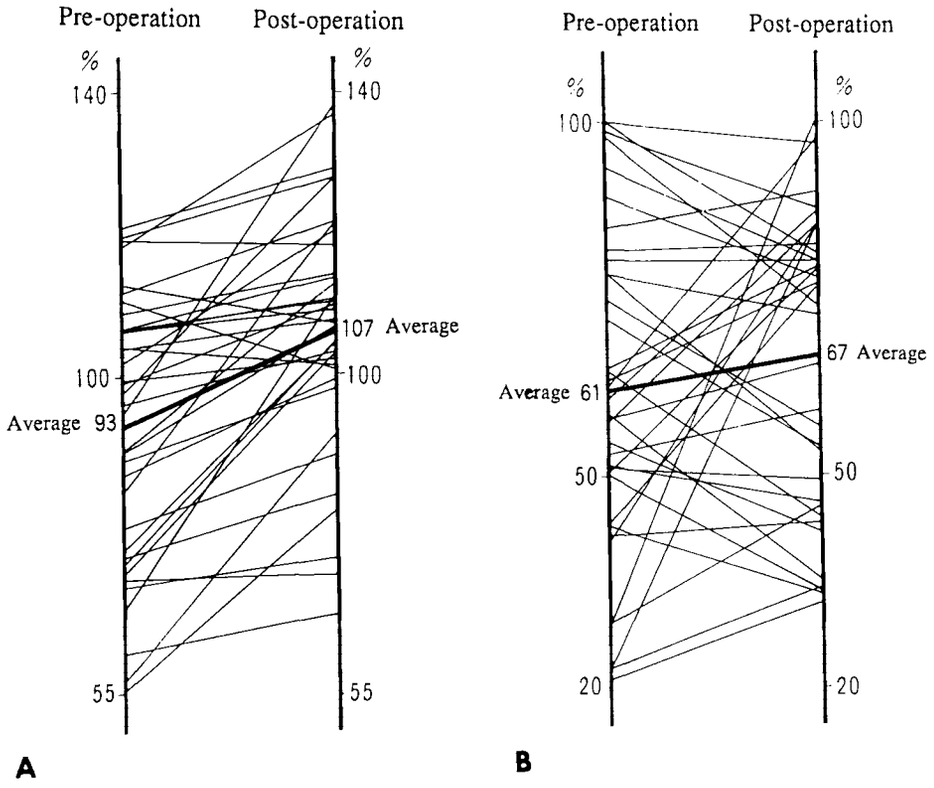


Fig 3. Preoperative and postoperative values for A) vital capacity and B) forced expiratory volume in 1 second.

increased upper airway resistance. But after seeing this increase in VC, we conclude instead that the most likely cause for the changes observed was an increased respiratory function due to the amplification of the VC after the surgery.[7,8]

There were patients who said that they could breathe better, and others who felt their throats were wider after the operation. The change in flow volume of FEV1 was from 60.6% to 66.7%. This indicates that the upper airway flow increased when the deviation of the larynx was corrected, and consequently the patients felt these effects. Only 8 cases (21%) were diagnosed as normal by autospirometer. Among the rest, 19 cases (50%) were diagnosed as obstructive, 5 (13%) as restrictive, and 6 (16%) as combined. This means that respiratory function is restricted in a high percentage of adults with ankyloglossia. Obstructive respiratory failure was the most frequent diagnosis in our study population, and the cause of obstruction was probably increased upper airway resistance. This resistance resulted mainly from deviation of the larynx.

*Improvement in Complaints*. There were both subjective and objective complaints preoperatively. Of the subjective complaints, many improved. Of the objective complaints, only incorrect articulation was

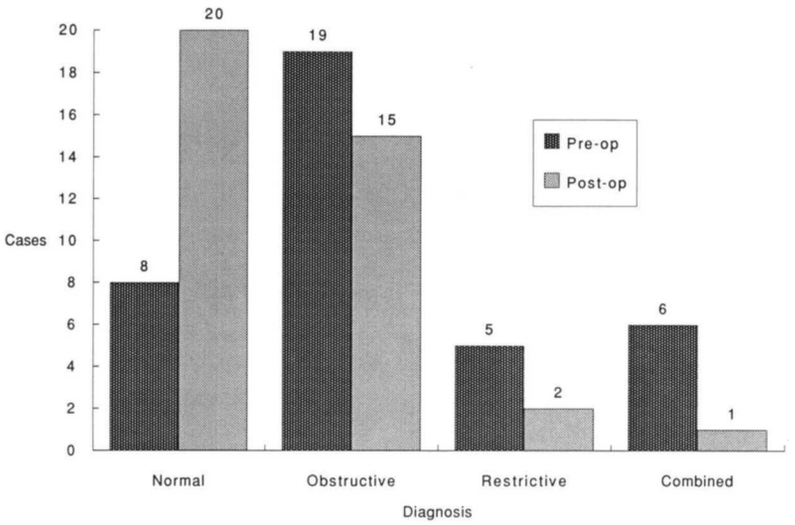


Fig 4. Changes in respiratory function as measured by Autospiro 500.

not affected; these patients did not realize their articulation was incorrect and therefore were not able to correct it.

Besides the 2 patients with noted muscle cramps, many other patients had muscle cramps during sleep. Muscle cramps occur frequently in pregnant women, in people with liver cirrhosis, and in old people, but the cause is unknown.[12-15] Our female patients had muscle cramps during pregnancy, too. We have reported that infants with ankyloglossia have pathologic muscle tonus.[1-6] We think ankyloglossia in adults also causes a state of pathologic muscle tonus that might develop as stiffness of the shoulders, muscle cramps, lumbago, snoring, fatigue, or respiratory failure.

There was a patient who could not play the flute well because tonguing was difficult. After surgery she could not only perform the tonguing well, but she also played with a more beautiful tone, with a narrowed glottic aperture.[16] Ankyloglossia with deviation of the epiglottis and larynx interferes with both tonguing and airflow in playing a wind instrument.

The patients' postoperative comments were, among others, that they had become calmer, that they had lost their feeling of anxiety and nervousness, that they were less irritable, and that their personalities had improved. The same tendency has also been observed in newborn, suckling, and postsuckling infants. It suggests that patients with ankyloglossia live in a state of stress.[17]

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