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Useful Genioplasty for Repeated Recurrent Sleep Apnea of Congenital Anomalies and Its Evaluation

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Background: Congenital facial anomalies with hypoplasia of the midface or lower face are associated with obstructive apnea syndrome. Although such patients underwent bone advancement surgery and their sleep apnea improved in the short term, it often recurred several years after surgery. It is difficult to perform another major osteotomy because of impairment of the facial contour or prior orthodontic treatment. Genioplasty was performed for genioglossus muscle advancement in patients with congenital anomalies and repeated sleep apnea. In this study, we evaluated the usefulness of this procedure and the mechanism for the improvement of sleep apnea.

Methods: Six patients were included: three with syndromic craniosynostosis, two with Treacher-Collins syndrome, and one with micrognathia by Goldenhar syndrome. Patients who had recurrence of sleep apnea after previous maxillomandibular osteotomies, or advancement and orthodontic treatment, received genioplasty for genioglossus muscle advancement. The patients were evaluated by body mass index, simple polysomnography, hyoid bone position on cephalogram, and the airway area on computed tomography images pre- and postoperatively.

Results: Polysomnography showed a significant improvement in the apnea-hypopnea index. Cephalometric measurement showed significant results of the hyoid bone position from point B and the ramus plane. However, no significant results were obtained in the airway area assessment.

Conclusions: Genioplasty for genioglossus muscle advancement can improve apnea-hypopnea index by moving the hyoid bone forward. Genioplasty was useful in patients with congenital anomalies who had a recurrence of sleep apnea after several procedures. *(Plast Reconstr Surg Glob Open 2023; 11:e4858; doi: 10.1097/GOX.0000000000004858; Published online 14 March 2023.)*

INTRODUCTION

Congenital anomalies of the face and hypoplasia of the midface such as syndromic craniosynostosis and hypoplasia of the lower face such as Treacher-Collins syndrome are associated with obstructive sleep apnea syndrome.[1,2] Although such patients underwent bone advancement surgery and their sleep apnea improved in the short term, it often recurred several years after surgery.[3] These patients had already undergone several major osteotomies and had completed orthodontic treatment. It is difficult to perform another major osteotomy for sleep apnea because it may change the facial contour radically or alter the occlusion. The treatment options for these patients are limited.

Reiley et al[4] and our group[5] reported the efficacy of mandibular osteotomy and hyoid bone advancement in obstructive sleep apnea. Genioplasty for genioglossus muscle advancement has been reported to improve the apnea-hypopnea index (AHI) in patients with obstructive sleep apnea syndrome.[6] Genioplasty is thought to be less invasive, is limited to the facial contour, and does not affect the occlusion. In this study, we performed genioplasty

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in patients with congenital anomalies such as syndromic craniosynostosis and Treacher-Collins syndrome who had repeated sleep apnea after surgery, and we performed several pre- and postoperative evaluations to determine the usefulness of the procedure and the mechanism for the improvement of sleep apnea.

PATIENTS AND METHODS

In this study, we examined six patients (two male and four female): three with syndromic craniosynostosis (two with Crouzon syndrome and one with Apert syndrome), two with Treacher-Collins syndrome who underwent genioplasty for obstructive sleep apnea, and one with micrognathia by Goldenhar syndrome who underwent genioplasty for closing the tracheostomy. Patients were treated at Osaka Medical and Pharmaceutical University from 1999 to 2021. The mean age of the patients who underwent genioplasty was 16 years (range, 10–23 years). Patients who had undergone previous osteotomy or bone advancement of the upper jaw or mandible, and those receiving orthodontic treatment had recurrent sleep apnea (Table 1).

A 3D model was created from the CT images and a preoperative simulation was performed. From the CT images, the distance to mental nerves and the root of the tooth was measured. Genioplasty was performed by osteotomizing the mandibular chin region in the shape of a baseball home plate to avoid injury to the canine teeth and damage to the mentonian nerve (Fig. 1).

The osteotomized segment was moved forward, with the musculature attached to the mental spine (Fig. 2). The amount of movement averaged 10 mm (7–13 mm), depending on the full thickness of the mandible. The anteriorly moved bone segment was then fixed using a titanium plate. In case 2 of Crouzon syndrome, genioplasty and distraction genioplasty were performed due to the recurrence of sleep apnea. During distraction, osteotomy was performed parallel to the occlusal plane in the shape of a home plate, as was done in the previous genioplasty, and an internal distraction device was attached to both the left and right sides (Fig. 3). The patient was evaluated before and after the two procedures (Figs. 1–3).

Takeaways

Question: Congenital anomaly patients underwent bone advancement surgery and their sleep apnea improved, but it often recurred several years after the surgery. We performed genioplasty for genioglossus muscle advancement in patients who had recurrent sleep apnea after the major osteotomy. This study aimed to evaluate the usefulness of the genioplasty and the mechanism for improvement of sleep apnea.

Findings: The polysomnography show a significant improvement in apnea hypopnea index. The cephalometric measurement showed significant results of the hyoid bone position from point B and ramus plane.

Meaning: Geniplasty was useful in patients with congenital anomalies who had recurrence of sleep apnea after several surgeries.

Clinical Symptoms

The patients were evaluated for pre- and postoperative changes of symptoms such as snoring, and sleep quality.

Polysomnography

Sleep apnea testing was performed using a simplified version of polysomnography. Patients were hospitalized overnight for testing. Sleep AHI, minimum SpO_2, and mean SpO_2 were measured, comparing the values pre- and postoperatively. The patients' body mass index (BMI) was evaluated pre- and postoperatively, and its relationship with the apnea index was examined.

Table 1. Patients on Whom We Performed a Genioplasty and Evaluated, with Descriptions of Orthodontic Treatment and Prior Surgical History

Patient No.	Gender	Diagnosis	Age(y)	Operation Method	Orthodontic Treatment	Previous Operation Age (y)/Operation Methods
1	Female	Apert syndrome	23	Genioplasty	Completed	4/Le Fort IV minus glabella advancement osteotomy
2	Male	Crouzon syndrome	17 23	Genioplasty distraction genioplasty	Completed	9/Le Fort III distraction osteotomy 14/Le Fort III advancement osteotomy + tongue reduction 15/Le Fort I advancement osteotomy + sagittal split ramus osteotomy 17/Sliding genioplasty
3	Female	Crouzon syndrome	10	Genioplasty	Not completed	8/Le Fort III advancement osteotomy
4	Male	Treacher-Collins syndrome	16	Genioplasty	Completed	10/Mandibular distraction osteotomy
5	Female	Treacher-Collins syndrome	23	Genioplasty	Completed	Mandibular segmental osteotomy (simultaneous enforcement)
6	Female	Micrognathia by Goldenhar syndrome	12	Genioplasty	Not completed	2/Tracheostomy 6/ Bilateral mandibular body distraction 8/ Reconstruction of the right temporomandibular joint a rib and costochondral complex graft

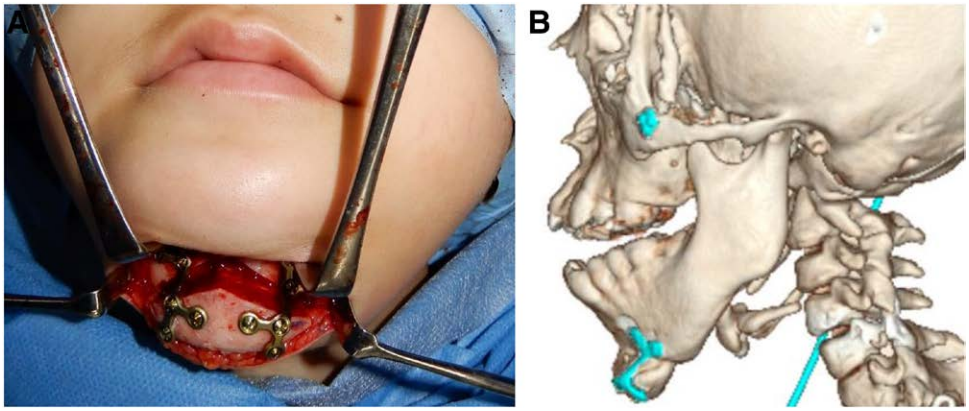

Fig. 1. The genioplasty for genioglossus muscle advancement in a 10-year-old girl with Crouzon syndrome (case 3). A, The mandible was osteotomized in the shape of a baseball home plate. The osteotomized segment was advanced 7 mm anteriorly with the muscles attached to the mental spine and fixed by mini plate systems. B, Three-dimensional CT scan image after the operation.

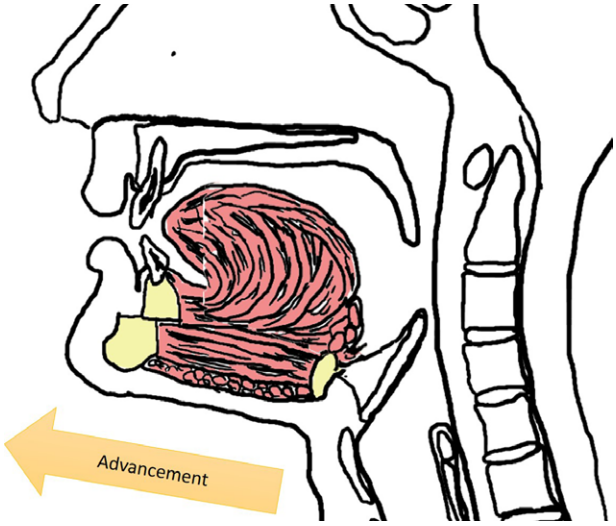

Fig. 2. The genioplasty. The osteotomized segment was moved forward with the musculature attached to the mental spine.

Cephalogram Evaluation

Preoperative and postoperative cephalograms were obtained and evaluated. To evaluate how the hyoid bone moved after genioplasty osteotomy, the shortest distance from the ramus plane to the hyoid bone and from the measurement points (point A, point B) were measured. The standard planes and points above were considered unchanged after genioplasty (Fig. 4).

CT Images Evaluation

Computed tomography (CT) images were obtained before and after surgery. Using Real INTAGE image analysis software, the evaluation was performed to measure the area of the airway using a plane parallel to the orbitomeatal line at the level of the uvula, epiglottis, and center of each cervical vertebra from C2/3 to C7/Th1, with the center point set to the dens axis (Figs. 5 and 6). All tests were performed at least 6 months postoperatively. In case 5, the patient was evaluated only with a simple polysomnography test because there were no data on the cephalogram and CT images. In case 6, the patient was evaluated using a cephalogram and CT images because the patient had undergone a tracheostomy at the age of 2 years, and a simple polysomnography test could not be performed (Figs. 5 and 6).

Statistical Analysis

All values were compared using pared *t* tests. The correlation coefficient between BMI and AHI was evaluated using Pearson's correlation coefficient. Differences were considered statistically significant at *P* less than 0.05.

RESULTS

Clinical Symptoms

In case 1 and case 2, the frequency of continuous positive airway pressure was decreased. In case 3, the snoring was decreased. In case 4, sleep quality was improved. In case 5, the body weight was increased 10 kg.

Polysomnography

In all patients, except one, the AHI of the sleep apnea test showed statistically significant improvement. Both the lowest and average SpO_2 in the test improved in all cases, but the overall difference was small and not statistically significant (Table 2).

Cephalogram Evaluation

Cephalometric measurements showed that the hyoid bone-point B distance showed significant shortening in all cases, and the hyoid bone-ramus plane distance was significantly prolonged in all cases (Table 3). The hyoid bone-point A distance was shortened in four cases and prolonged in two cases, with no significant difference.

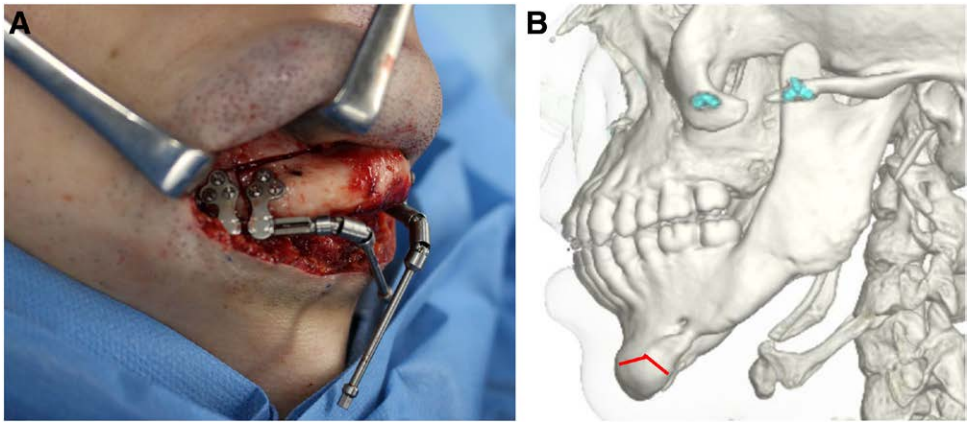

Fig. 3. The genioplasty for genioglossus muscle advancement by distraction osteogenesis in a 23- year-old man with Crouzon syndrome (case 2). A, The mandible was osteotomized in the shape of a baseball home plate. The internal distraction devices were fixed to both sides. B, The preoperative 3D CT image. The red line shows the osteotomy line.

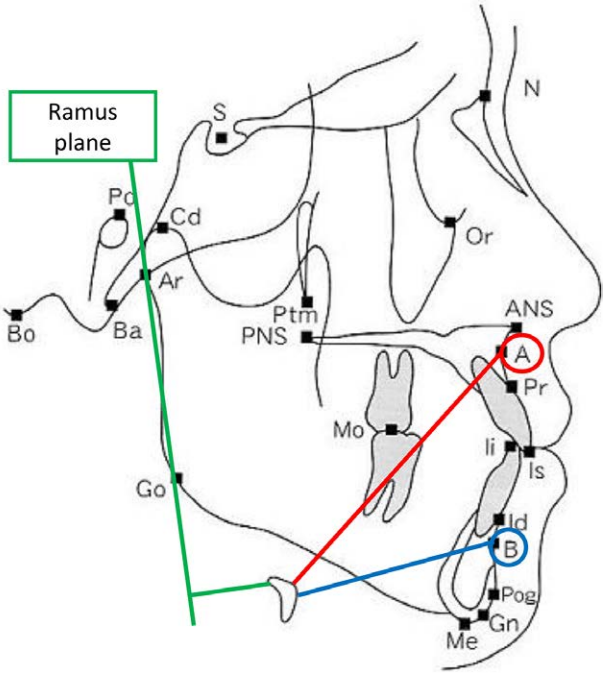

Fig. 4. The landmarks used in evaluation of the cephalogram. The shortest distances from points A, B and the ramus plane to the hyoid bone were measured.

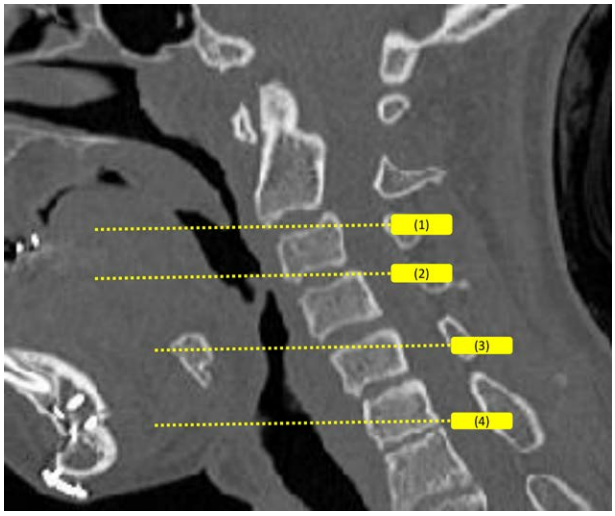

Fig. 5. The levels which indicate the sites of measurement of the airway area. The yellow dotted lines are parallel to the orbito-meatal line and show the lines at the height of (1) the uvula, (2) the epiglottis, (3) the center between each cervical vertebra, and (4) the center point of each cervical vertebra.

CT Image Evaluation

The results of the airway area measurements were not consistent, with some cases showing enlargement and some showing reduction (Figs. 7–9). The hyoid area, which might be expected to be the most enlarged, was actually enlarged in case 2 with Crouzon syndrome and case 6 with micrognathia by Goldenhar syndrome, whereas it was slightly reduced in the other cases. The results showed enlargement of the pediatric patients several years postoperatively (Figs. 7–9).

BMI and Apnea Index

In case 1 with Apert syndrome, the BMI seemed to be related to the AHI; therefore, the AHI worsened as the BMI increased. (**See figure, Supplemental Digital Content 1,** which displays the relationship between BMI and AHI in cases 1 to 4. **http://links.lww.com/PRSGO/C444**.) In a study of the association between AHI and BMI, a positive association was found only in case 1, but no association was found in the other cases. **(SDC1, http://links.lww.com/PRSGO/C444)**.

AHI was related to an operation episode. In all cases except case 1, the AHI decreased markedly immediately after the operation **(Supplemental Digital Content 1, http://links.lww.com/PRSGO/C444)**.

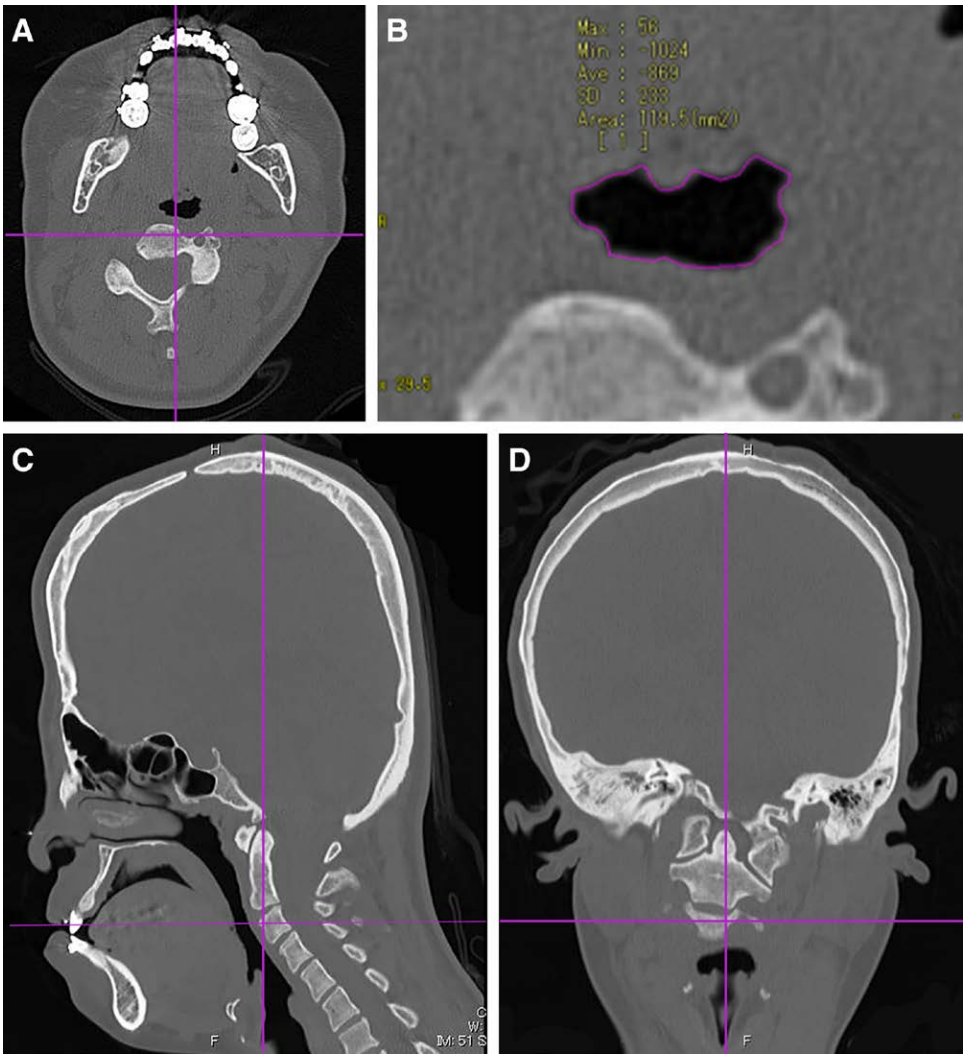

Fig. 6. The schemas show the images of CT scan and indicate the airway area measurement method. A, The slice which is parallel to the OM line. B, Magnified image of the upper right and indication of the airway area that should be measured using Real INTAGE. C-D, Images showing that the center point of the measuring plane was set to the dens axis.

DISCUSSION

Congenital anomalies of the face, such as hypoplasia of the midface or lower face, are associated with obstructive sleep apnea syndrome.[4,6] Patel and Fearon reported that 10 of 32 patients with syndromic craniosynostosis presented with obstructive sleep apnea preoperatively, eight (80%) normalized after distraction, and two patients did not completely normalize.[3] Two patients (20%) developed recurrent obstructive sleep apnea, and one patient developed de novo obstructive sleep apnea.[3] They discussed the need for continued vigilance for ventilatory impairment following midfacial advancement in growing children. It is commonly recognized that most children with syndromic synostosis will require more than one midfacial advancement over a lifetime.[3]

Because patients with syndromic craniosynostosis or other congenital anomalies with repeated recurrent sleep apnea have already undergone major osteotomy several times and have completed orthodontic treatment, it is difficult to perform another major osteotomy because of impairment of occlusion. Surgery for OSA is classified into phase I soft tissue surgery and phase II skeletal surgery. The phase I procedures include uvulopalatopharyngoplasty, hyoid suspension, and genioglossus advancement, which means a circular osteotomy to capture the genial tubercle. The phase I surgery was reported not to be sufficient to produce a long-term treatment.[7] Genioplasty was used as the final operative method for patients with congenital anomalies and recurrent sleep apnea.

The method of traction with the hyoid muscle group using genioplasty osteotomy was first reported by Riley et al.[4,6] This method is relatively less invasive and does not affect occlusion, making it suitable for patients without maxillary hypoplasia or those with an established occlusion, such as those who have completed orthodontic

Table 2. Results of the Simple Polysomnography before and after the Genioplasty

	AHI (Episodes/h)*		Lowest SpO_2 (%)		Average SpO_2 (%)	
Patient No.	**Before**	**After**	**Before**	**After**	**Before**	**After**
1	14.8	15.6	84	88	96	98
2 (first operation)	28.2	14.7	81	65	93	88
2 (second operation)	23.7	6.9	69	87	96	96
3	24.8	15	76	80	95	95
4	49.6	13.7	71	86	96	96
5	25.1	10.3	53	79		

*A significant result ($P < 0.05$).

Table 3. Results of the Cephalometric Analysis before and after the Genioplasty

	Point A		Point B*		Ramus plane*	
Patint No.	**Before**	**After**	**Before**	**After**	**Before**	**After**
1	91.24	93.19	60.05	55.59	3.23	10.48
2 (first operation)	71.59	69.3	45.34	39.15	6.37	10.29
2 (second operation)	71.9	65.24	36.51	33.87	12.01	17.19
3	82.61	76.94	43.01	40.1	6.1	9.31
4	84.66	80.87	44.64	42.79	1.76	2.43
6	71.95	79.02	36.06	30.79	5.11	9.18

*A significant result ($P < 0.05$).

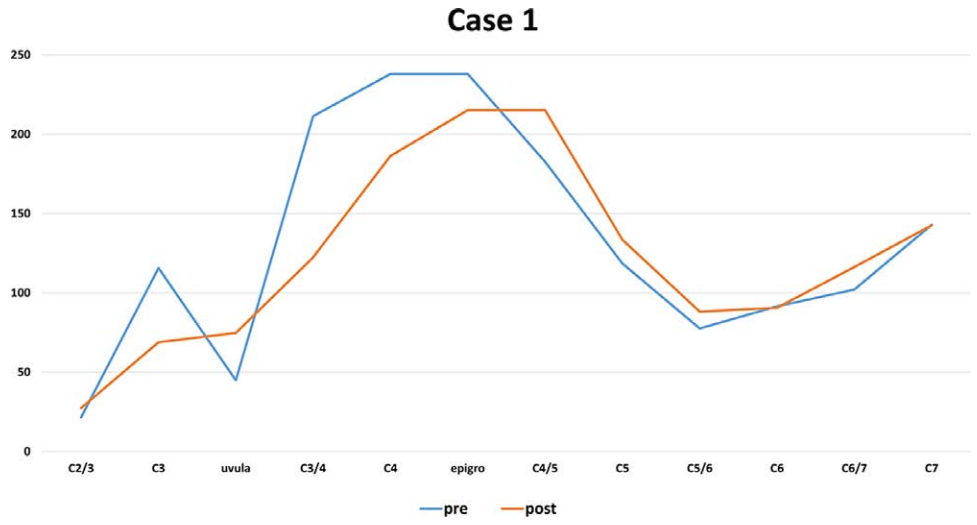

Fig. 7. The changes of the airway area in case 1 before and after the surgery. A slight airway enlargement was observed around the hyoid bone level. The hyoid bone level is between the C5/6 and C6 level.

treatment. Previously, we reported that horizontal and segmental osteotomies of the mandible in Treacher-Collins syndrome patients with obstructive sleep apnea syndrome improved sleep apnea.[5]

Genioplasty for genioglossus muscle advancement improves the AHI in patients with obstructive sleep apnea syndrome.[8] The concept of this method is the same as ours in this study, but the osteotomy method is different. Our method has resulted in improvement of the sleep apnea index and sleep quality, weight gain, and growth in those who underwent surgery in childhood, indicating that this technique is useful for patients who have undergone multiple procedures in the past and for those who have completed orthodontic correction. We adopted external incisions for the genioplasty because we wanted to perform safe osteotomy, checking the mental nerves and muscle attachment under direct vision. However, an intraoral incision should be adopted. For safe osteotomy, the use of a cutting guide has been reported.[9–11] The risk of nerve injury was reported to reduce to 2.5% at a safe distance of 7.06 mm from the inferior border of the mental foramen.[11]

To date, several studies have examined osteotomy evaluation methods and how they yield airway expansion; however, we focused on four points in this study. We discuss each of these as follows.

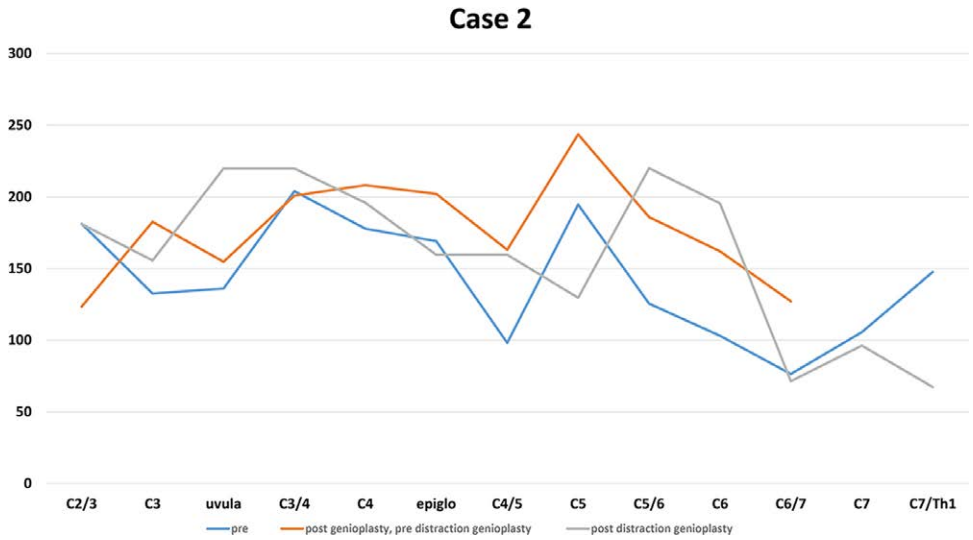

Fig. 8. The changes of the airway area in case 2. The first genioplasty was bone segment advancement and fixation. The second genioplasty was osteotomy and distraction. The airway enlargement was recognized after the first genioplasty and again after the distraction genioplasty. The hyoid bone level is defined as that of the C6.

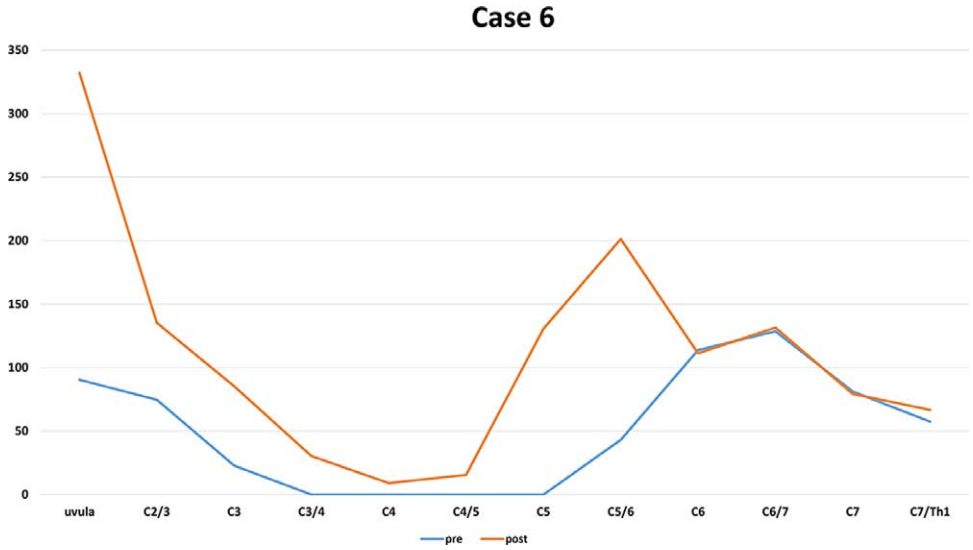

Fig. 9. The changes of the airway area in case 6. Comparatively, the airway area was enlarged in the upper airway. The hyoid bone level is defined as that of C5/6.

Polysomnography

AHI was significant in the sleep apnea test, and the lowest SpO_2 and average SpO_2 improved in all cases, indicating that genioplasty with genioglossus muscle advancement led to improvement of obstructive sleep apnea. These results show that genioplasty was effective as the final operative method for repeated recurrent sleep apnea due to congenital anomalies.

BMI and AHI

Only the patient in case 1 showed a correlation coefficient between AHI and BMI. No other patients showed any correlation. The difference between case 1 and the others is that case 1 has almost all the data for an adult, whereas the others have data for patients in their growth period. Adult patients have completed the growth of the skeleton around the airway. AHI is thought to increase as a result of large weight gain. This indicates that the amount of fatty tissue due to weight gain may narrow the airway. However, patients who are still in their growth period are thought to have the possibility that airway morphology will change with their growth.

Cephalogram

In a report by Fumino et al,[12] hyoid bone movement was measured in patients with maxillary prognathism who underwent genioplasty osteotomy, and they

reported a statistically significant forward movement of the hyoid bone. In our method, the reference points, points A and B, were originally anterior to the hyoid bone, and the distance from point B was significantly shortened in all cases. All cases showed an extension of the distance from the ramus plane to the hyoid bone, with significant results. This suggested that the hyoid bone moved anteriorly. These results revealed that genioplasty played an important role in decreasing obstructive sleep apnea by pulling the hyoid bone forward. It is reported that AHI correlated with the movement of the hyoid bone to the oral side.[13,14] Our results showed that the hyoid bone moved significantly forward and that AHI was improved significantly. We grouped all patients into the same category of morphologic abnormality, but we believe that the hyoid bone movement by the genioplasty improved the AHI to all categories of morphologic abnormality.

Airway Area

Flores et al reported that Le Fort III distraction in syndromic craniosynostosis significantly increased the airway in a cephalometric study.[15] Nout et al evaluated upper airway volume after Le Fort III advancement in patients with syndromic craniosynostosis and found a significant increase in volume but no correlation with the amount of movement.[16] Haisong et al reported a three-dimensional evaluation of airway morphology after Lefort III distraction in patients with syndromic craniosynostosis with obstructive sleep apnea syndrome and that volume improvement was achieved in the upper airway.[17] Maxillomandibular advancement has also been reported to increase the airway for obstructive sleep apnea.[18,19] Conversely, setback osteotomy of the mandible or two-jaws has been reported to decrease the pharyngeal airway.[20] Mandibular distraction of micrognathia has been reported to increase upper airway volume.[21]

Our results showed that the hyoid bone moved significantly forward, but there was no significant enlargement of the airway. Genioplasty may not result in significant changes in the upper airway because only part of the mandible is moved. In addition, a two-dimensional evaluation may be consistent because the airway morphology changes depending on the position of the tongue and the neck angle during the examination. Although no significant change in the area was observed, it is possible that the three-dimensional morphology itself may have changed. We found that the pediatric patients showed improvement in growth with long-term follow-up, and we believe that airway expansion is likely to be achieved as they grow.

CONCLUSIONS

Genioplasty osteotomy can improve the AHI by moving the hyoid bone forward. Genioplasty osteotomy in patients with congenital anomalies who had recurrent obstructive sleep apnea syndrome was useful because it was relatively noninvasive and improved sleep apnea attacks without altering occlusion.

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