

general health, within the context of identification of suspected COVID-19 patients primarily when they are asymptomatic.³⁶ A similar method can be used to help identify dental patients with possible long-term complications. Recovering COVID-19 patients may require liver function tests, kidney function tests, coagulation profile namely prothrombin time, and blood sugar level, as part of the treatment planning. Maxillofacial surgeons should consider consultation with patients' physicians if sufficient data pertaining to the medical history was not completely provided by the patient.

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Surgical Treatment of Oromandibular Limb Hypogenesis Syndrome Type I A by Distraction Osteogenesis Combined With Orthodontic Rehabilitation

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Abstract: Oromandibular limb hypogenesis syndrome is a rare developmental anomaly and only a few cases are reported with complete surgical and orthodontic rehabilitation. An adult male patient with isolated hypoglossia, micrognathism, hypodontia, (oromandibular limb hypogenesis syndrome type I A) was treated

with a combination of distraction osteogenesis and orthodontic intervention. The patient was followed up for the duration of 6 years from his first visit to 4 years after the surgery. The combined procedure resulted in successful and satisfactory treatment of the patient by restoring facial aesthetics, occlusal balance, and functional harmony. However, there was not enough tongue enlargement due to late surgical intervention. The objective of this report is to describe the etiology of hypoglossia, the consequences for oral function, and to share our experience from the oral rehabilitation during the treatment procedure.

Key Words: Distraction osteogenesis, hypoglossia, micrognathism, orthodontics, rehabilitation

Hypoglossia is a rare congenital abnormality. Hypoglossia can occur either as an isolated malformation or in association with other deformity.¹ This anomaly is almost associated with the malformation in the upper and the lower extremities and dental agenesis. However, isolated hypoglossia is even rarer. As a consequence of the lack of muscular stimulus between the alveolar arches, these do not develop transversely and the mandible does not grow in the anterior direction, resulting in severe transverse deficiency of the mandible, hypodontia, anterior crowding, and unilateral or bilateral maxillomandibular discrepancies.²

The prevalence of isolated hypoglossia is unknown and appears to be very rare. At the time of this writing, there are very few cases of hypoglossia reported.¹ In cases of isolated hypoglossia and micrognathism distraction osteogenesis (DO) and orthodontic intervention are the mainstay of treatment.^{3,4} Recent studies report that the mandibular widening by DO followed by orthodontics is an effective treatment for mandibular transverse deficiency and associated telescopic bite.⁴ A 6-year follow-up case of oromandibular limb hypogenesis syndrome type I A treated combine with DO and orthodontic rehabilitation is reported.

CLINICAL EXAMINATION

Extraoral examination revealed a convex facial profile and reduced lower anterior facial height at the frontal view. Deep labiomental

fold accompanied mandibular retrognathism which showed the short facial type and shallow mandibular plane.

The maxilla and the mandible both were taper arch form (V-shaped) with maxilla being narrow and high palatal vault. There was severe anterior crowding in both maxilla and the mandible and 31 (mandibular left central incisor), 41 (mandibular right central incisor), and 42 (mandibular right lateral incisor) were missing. He had micrognathism and hypoglossia without limb anomalies. He had no history of any trauma and not any significant family history. Moreover, he had no other abnormal physical nor mental history contraindicating treatment. The extraoral photographs, radiographs, and intraoral photographs of the patient are shown in Figures 1–3 respectively.

TREATMENT PROGRESS AND RESULTS

The surgery was performed when the patient was 18 years old. Three distractors were placed on both sides of the body of the mandible, and the mandibular midline distractor on the symphysis area for the lengthening and widening of the mandible. An intraoral DO device was used to avoid facial scarring, pin tract infections, and high visibility. At the same time, the midpalatal suture was opened surgically and tooth-borne rapid maxillary expansion was placed on the maxilla and expanded.

After a sufficient maxillary expansion, extraction of 14 (maxillary right first premolar) and 24 (maxillary left first premolar) was done to correct the overjet and the space created was closed by fixed orthodontic appliance for 16 months. Endodontic treatment was done in 32 and was restored by post and core treatment followed by a crown. The patient's mandible grown significantly, the occlusion relationship is normal, and the appearance is better. The overall treatment process was 45 months.

DISCUSSION

The exact cause of hypoglossia is difficult to understand. In hypoglossia, the tongue has a significantly reduced volume which leads to an unbalanced equilibrium, resulting in lingual inclination of the teeth due to the outer pressure from the circum-oral muscles. Therefore, under this condition correcting malocclusion is a

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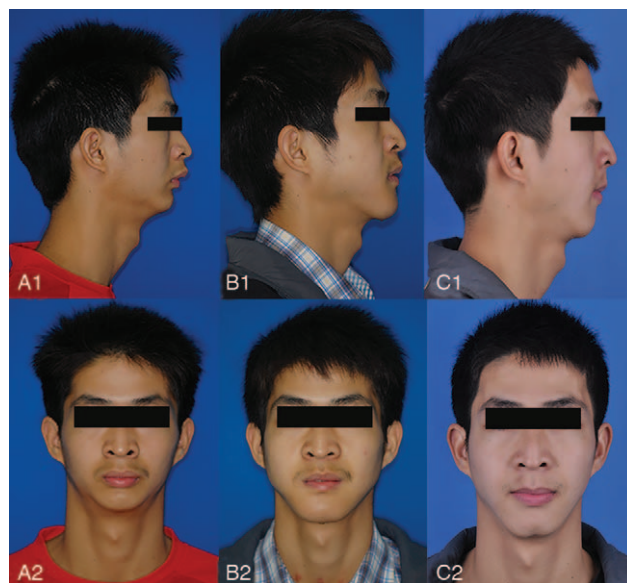


FIGURE 1. Extraoral photographs, A1 and A2: presurgery lateral and frontal profile, B1 and B2: 3 months postsurgery lateral and frontal profile, C1 and C2: posttreatment lateral and frontal profile, respectively.



FIGURE 2. Intraoral photographs, A1, A2, and A3: presurgery maxillo-mandibular relation, maxillary dentition, and mandibular dentition, B1, B2, and B3: 3 months postsurgery maxillo-mandibular relation, maxillary dentition, and mandibular dentition, C1, C2, and C3: posttreatment maxillo-mandibular relation, maxillary dentition, and mandibular dentition, respectively.

great challenge. It also has a great possibility of relapse in the long-term.

Surgically assisted maxillary expansion has been recommended as a solution for transverse problems in adult patients.⁵ Distraction osteogenesis at both sides of the mandible and mandibular midline distraction can improve the skeleton and soft tissue profile, as well as reduce the degree of relapsing.⁶

In this case, the expansion of the midpalatal suture and the DO of the mandible were performed at the same time. One of the main challenges during the distraction process is controlling the direction of the newly formed bone. During DO, forward advancement of the mandible resulted in premature occlusal contact of the posterior teeth, which lead to clockwise rotation of the mandible. Therefore, as the mandible lengthened, anterior open bite and an increased lower facial third were noted. In postoperative orthodontic treatment, it is necessary to properly intrude the posterior teeth and extend the anterior teeth to improve the vertical bite. The study of Strijen et al⁷ showed that high-angle patients have a higher risk of recurrence after DO.

During lengthening of the mandible, the condyle may have moved backward and caused stress on the condyle surface.⁸ In this study, the length of the ramus was shorter than pretreatment due to the counterforce created by the distractors upon the condyle, which lead to the backward and clockwise rotation of the condyle resulting in backward and upward movement of the Gonion point.

Many studies reported no incidence of temporomandibular joint pain following widening of the mandible.⁹ Some studies have reported patient with temporomandibular pain and dysfunction after mandibular widening. However, in our case the patient has not reported any signs of temporomandibular dysfunction.

In mandibular symphysis distraction, osteotomy must be carried out with great care to avoid encroaching on or injuring the periodontal ligaments adjacent to the osteotomy line. If the osteotomy line is encroached by periodontal ligament tissues, the distraction gap fills with bone and moderate amounts of periodontal ligament-like tissues, leading to pseudo-union. In our case report, the lower anterior region was severely crowded, thus causing difficulty in performing osteotomy. However, DO was performed and obtained good results after consolidation.

There is no evidence that the expression of this equilibrium has an effect on the stability of lower dentition after mandibular widening. However, the dental arch can theoretically appear when there is equilibrium between buccal pressure and tongue pressure. However, in this case, due to hypoglossia, prolonged retention was

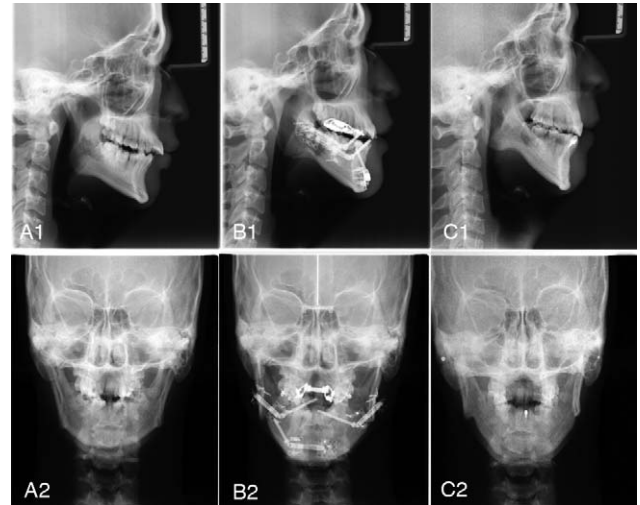


FIGURE 3. Radiographs, A1 and A2: presurgery lateral and frontal cephalogram, B1 and B2: 3 months postsurgery lateral and frontal cephalogram, C1 and C2: posttreatment lateral and frontal cephalogram, respectively.

necessary. As the mandible widened and lengthened, the mandible increased in volume and occlusal function improved.

CONCLUSIONS

In conclusion, early surgical correction is necessary for better occlusal function and speech. If treatment of hypoglossal patients with midline mandibular distraction is performed during the growing age, it could help to regain the normal tongue size spontaneously. Long-term retention was necessary to reduce the risk for transversal relapse. The combined orthodontic-orthognathic surgical procedure helps to improve the quality of life in patients with hypoglossia with transversal discrepancies.

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