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CASE REPORT

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Treatment of severe micrognathia in an adult with distraction osteogenesis: A case report

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Key Clinical Message: Distraction osteogenesis (DO) of the mandible is often performed at a young age, and there are few reports after age 30, as in this case. The Hybrid MMF used in this case was useful in that it allowed correction of fine directionality.

Abstract: DO is often performed in young patients with a high capability of osteogenesis. We performed distraction surgery for a 35-year-old man who had severe micrognathia with serious sleep apnea syndrome. Four years postoperatively, suitable occlusion and improvement of apnea were observed.

KEYWORDS
distraction osteogenesis, mandible, micrognathia, occlusal guidance, sleep apnea syndrome

1 | INTRODUCTION

Micrognathia usually occurs due to congenital craniofacial anomalies, including Pierre Robin sequence, Treacher Collins syndrome, and hemifacial microsomia. Airway obstruction in the neonatal period caused by severe micrognathia requires early treatment.[1] In infants with micrognathia due to Pierre Robin sequence, management of airway obstruction may require tracheostomy.[2] Białobrzeska et al. reported that distraction osteogenesis (DO) in early infancy is an alternative to avoid tracheostomy. Even if there are no breathing problems when awake, the patient may have airway obstruction during sleep. In addition to airway obstruction, micrognathia causes masticatory and speech disorders due to occlusal insufficiency. Moreover, since facial aesthetics are severely compromised, skeletal improvement is required.[1] Many studies have reported on surgery for micrognathia and the application of DO for severe micrognathia.[3] However, surgical treatment for micrognathia is usually indicated for younger patients because preoperative treatment, such as orthodontic treatment, is often started at an early age and is completed by adulthood. Hence, DO is mainly performed in teenagers or young adults.[4]

We report the case of a patient with mandibular micrognathia and sleep apnea who underwent DO.

2 | CASE HISTORY

A 35-year-old man was referred to our department with a chief complaint of micrognathia. The patient had no specific general history, and facial examination revealed severe mandibular retrognathia without asymmetry (Figure 1A,B). No other facial morphological abnormalities (e.g., auricular deformities) were observed. Intraoral examination revealed labial inclination of the upper and lower anterior teeth. An open bite was observed from the right to the left canine with an overbite of −7 mm and an overjet of 10 mm (Figure 1C,D). A panoramic radiograph showed hypoplasia of the bilateral mandibular condyles, and the condyles deviated from the mandibular fossae (Figure 1E). Cephalometric radiographs showed severe upper airway stenosis (Figure 1F), and computed tomography showed hypoplasia of the bilateral mandibular condyles and shortening of the rami. The patient's Apnea Hypopnea Index (AHI) was 62.9, which was a therapeutic indication for sleep apnea syndrome (SAS). Orthognathic treatment with mandibular distraction was planned to improve aesthetics and treat SAS.

Owing to the incisor inclination, the amount of mandibular advancement required was >20 mm.

Approximately 1 year after the initiation of orthodontic treatment, an extension device was surgically implanted. A skin incision was made in the submandibular region and the mandible was visualized. An osteotomy was performed at the planned site using an ultrasonic bone scalpel. Mandibular Zurich Pediatric Ramus Distractors (25 mm advancement type, KLS Martin) were fitted so that the direction of mandibular extension and maxillary occlusal plane were as parallel as possible. A rotation screw rod was placed outside because the oral cavity was very narrow. No incision was made intraorally. After 5 days of rest, distraction was started by rotating the device twice a day to achieve 1 mm distraction. A mandibular extension of 25 mm was completed in 25 days. During the distraction period, a Smart Lock Hybrid Maxillomandibular Fixation (Hybrid MMF) device (Stryker) was used for rigid fixation because of the high risk of tooth extrusion when using a multi-bracket appliance for occlusal guidance. Because the Hybrid MMF has many ridges, elastic was used to adjust the direction of distraction. No signs of infection or severe pain were observed during distraction (Figure 2). One month after DO, the rotation screw rod was cut and the skin wound was sutured. Occlusal guidance was continued for 6 months using Hybrid MMF. Six months postoperatively, the sleep apnea test was repeated, and the AHI decreased to 6.5. The distraction device was removed 1 year postoperatively because there were no signs of relapse or change in molar occlusion. Simultaneously, a genioplasty was performed to improve aesthetics. The distracted area was completely ossified. No changes in the condylar position or signs of relapse were observed 4 years after distraction (Figure 3).

3 | DISCUSSION

Micrognathia is one of the most common congenital facial deformities and is the main sign of Pierre Robin sequence.[1] Extremely small mandibles are easily diagnosed visually soon after birth, and treatment may be initiated early.[2] However, in the present case, no syndrome or other disorder related to micrognathia was diagnosed in childhood. The patient was short (148 cm) and underweight (45 kg), compared with the average adult Japanese male (age, 30–39), whose height and weight, according to the

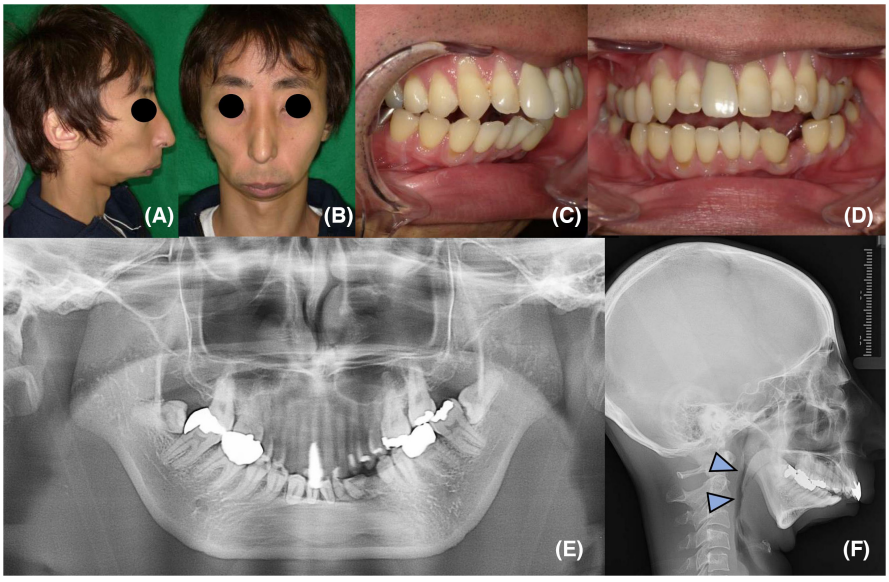

FIGURE 1 Findings at the initial visit. (A, B) Extraoral photographs (facial view): Severe mandibular retrognathia. (C, D) Intraoral photograph: An open bite. (E) Panoramic radiograph: Hypoplasia of the bilateral mandibular condyles and deviation of the condyle from the mandibular fossa. (F) Cephalometric radiograph: Severe upper airway stenosis.

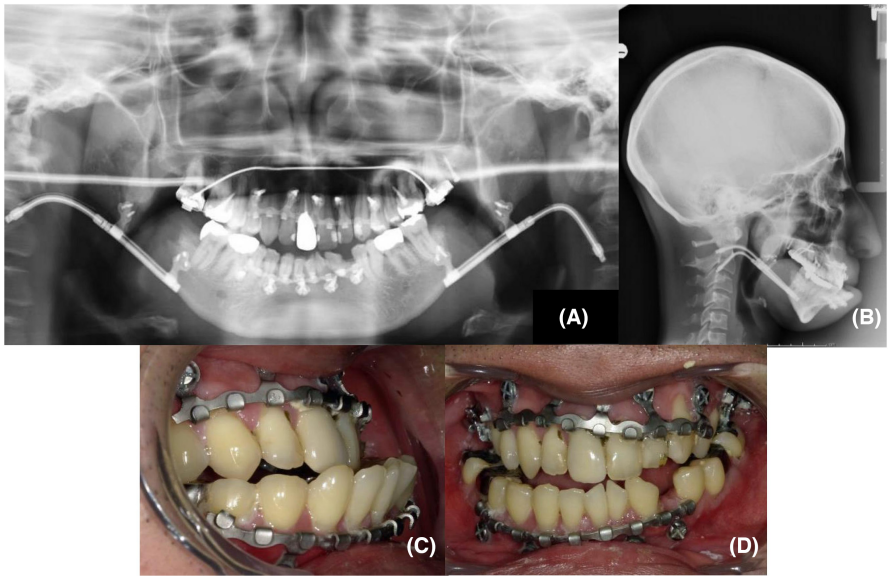

FIGURE 2 Attached mandibular distraction and Hybrid MMF. (A) Panoramic radiograph: Immediately after DO, no obvious osteosclerotic lesions. (B) Cephalometric radiograph: The mandible is moved anteriorly. (C, D) Intraoral photographs: Hybrid MMF attached.

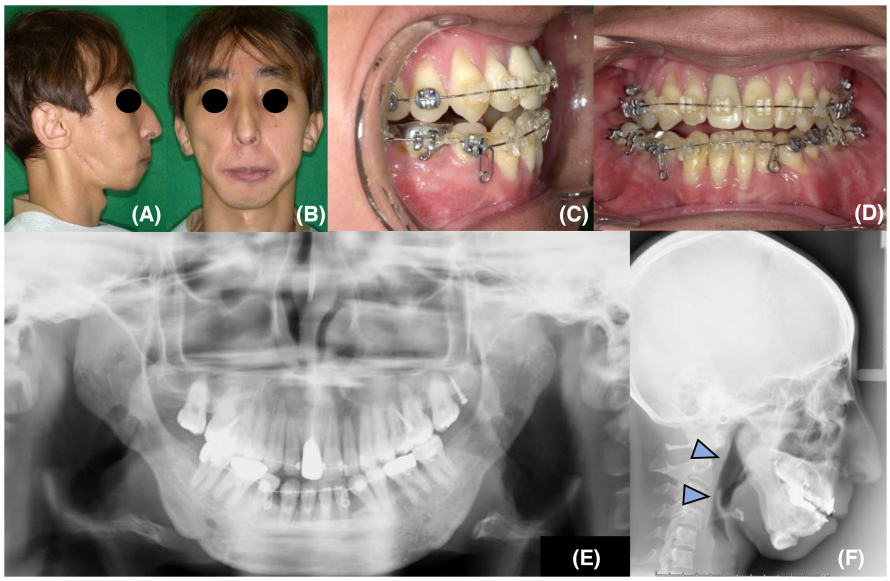

FIGURE 3 Findings 4 years after distraction. (A, B) Extraoral photographs (facial view): Mandibular retrognathia has been corrected. (C, D): Intraoral photograph: No signs of relapse or change in molar occlusion. (E) Panoramic radiograph: Osteosclerotic lesions are observed. (F) Cephalometric radiograph: Upper airway stenosis has been resolved.

National Health and Nutrition Survey (2018), Ministry of Health, Labor and Welfare, Japan, are 172.2 ± 6.1 cm and 71.0 ± 12.3 kg, respectively. Therefore, this very severe micrognathia could most likely be an effect of some systemic disease.

DO was introduced in the 1960s and was used for orthopedic surgery, such as in short-limbed dwarfism.[5] McCarthy et al. first applied DO in dentistry in 1992 to lengthen mandibles in children.[6] Since then, many orofacial cases have been reported. DO has been used to alleviate mandibular hypoplasia and asymmetry, hemifacial microsomia, Goldenhar's syndrome, maxillary arch-width discrepancy, and severe midface deficiency.[7–9] Mandibular retrognathia leads to glossoptosis, airway obstruction, and obstructive SAS at an early age. Some children with severe airway obstruction need early mandibular advancement.[1,2,10,11] Severe SAS was present in this case, and significant improvement was achieved with DO. Thus, DO is effective in treating SAS not only in young patients but also in adults.

DO is often performed in children, teenagers, and young adults, who have a high ability for osteogenesis.[1,12] Although DO in adults has been reported,[4,12–17] only two patients were aged >30 years: a 65-year-old man with unilateral distraction[16] and a 34-year-old man with bilateral mandibular DO using an intraoral tooth-borne device.[4] The small number of adult cases might be due to the concern regarding whether sufficient bone would be formed by DO, and because DO is usually performed at a younger age, as treatment for micrognathia begins in childhood. However, it was possible to perform a 25-mm-long DO even after 35 years of age. To the best of our knowledge,

this is the first case of bilateral mandibular DO in a patient aged >35 years.

DO has some limitations, including infection of the surgical site and distractor, long hospitalization, and relapse.[18] A high mandibular-plane angle is reported to be the major etiological factor contributing to postoperative skeletal relapse.[19] Larger advancements (>10 mm) are associated with an increased risk of relapse due to perimandibular soft-tissue tension.[20–23] In contrast, DO has been reported to have a lower incidence of relapse, bone compression, and bone resorption than osteotomy, which involves a single bone movement. Moreover, many studies have reported that mandibular advancement ≥10 mm is possible using DO accompanied by simultaneous expansion of the functional soft tissue matrix, including blood vessels, nerves, muscles, skin, mucosa, fascia, ligaments, cartilage, and periosteum.[19,20,24] In our case, we determined a high risk of relapse after mandibular advancement using sagittal split ramus osteotomy because of condylar hypoplasia, deviation from the mandibular fossa, and advancement ≥20 mm. A previous study reported that relapse after DO is likely to occur within 6 months postoperatively.[25] In this case, no relapse of the mandibular bone occurred after 4 years postoperatively, and good occlusion and aesthetics were achieved.

Before performing orthognathic surgery, the amount of movement is commonly examined using preoperative simulation, and orthodontic treatment is performed first to stabilize the occlusion.[26] However, it is difficult to accurately predict the final occlusal position in DO due to the direction of occlusal guidance and the possibility of retroversion. Therefore, a technique that allows extension to the exact planned position as far as possible must be used.

Occlusal guidance is usually performed using a multibracket orthodontic appliance with intermaxillary elastics, and a fixation source is often sought on the teeth.[27] However, in teeth with periodontitis, strongly tilted axes, or extrusion, using the tooth as a fixation source for occlusal guidance causes extrusion, which exacerbates the tooth condition.[27] An intermaxillary fixation screw is frequently used to avoid these complications.[28] However, since a large number of intermaxillary fixation screws cannot be implanted, the direction of guidance is also limited. In this case, Hybrid MMF was selected as a device that would not put a burden on the teeth and would allow detailed adjustment of the guidance direction. Particularly in this case, we checked the attachment site and strength of the elastics daily and attempted to extend the mandible in the appropriate direction. Consequently, no dislocation or posterior deviation of the condylar head was observed.

4 | CONCLUSIONS

We performed a distraction surgery for a 35-year-old man with severe micrognathia and serious SAS. Four years postoperatively, a favorable occlusion and improvement of apnea were observed. Even at 35 years of age, DO as long as 25 mm was possible. Hybrid MMF was useful in allowing fine directional correction and long duration of stability in DO.

AUTHOR CONTRIBUTIONS

Katsuhisa Sekido: Resources; writing – original draft. **KUMIKO FUJIWARA:** Resources; supervision; writing – review and editing. **Hidetake Tachinami:** Visualization. **Shuichi Imaue:** Visualization; writing – review and editing. **Keishi Hanashiro:** Resources. **Makoto Noguchi:** Supervision.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest associated with this manuscript.

DATA AVAILABILITY STATEMENT

All data generated or analyzed during this study are included in the published article.

CONSENT

Written informed consent was obtained from the patient for the publication of this case report including all patient data.

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