The present case report describes the combination of orthodontic treatment and two-stage surgery used to treat a 16-year-old female with a severe maxillary occlusal cant and a mandibular deviation. The patient was diagnosed with right temporomandibular joint ankylosis six months after suffering a facial injury at five years of age. A unilateral condylectomy was performed 12 months later. A hybrid functional appliance was used between nine and 16 years of age to prevent deterioration of the maxillary occlusal cant. At 16 years and six months, it was determined that the patient required a combination of surgical and orthodontic treatment, the pre-surgical orthodontic phase of which was completed at 19 years. The surgery was performed in two stages, which involved a Le-Fort 1 procedure to intrude the maxilla in the left molar region and to extrude in the right molar region. Distraction osteogenesis was performed to achieve a lengthening of the right mandibular ramus. The second operation was a unilateral sagittal split ramus osteotomy to achieve a forward and upward rotation of the left hemimandible. As a result, the patient's facial asymmetry and occlusion significantly improved. The improvements were well maintained and the patient remained stable after one year of retention.

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Orthodontic treatment in conjunction with distraction osteogenesis and orthognathic surgery for severe facial asymmetry

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Introduction
A condylectomy to repair a unilaterally ankylosed temporomandibular joint (TMJ) during the growth period often results in a maxillary occlusal cant and accompanying mandibular deviation. The maxillary occlusal cant may worsen due to asymmetrical jaw growth.1,2 Combined surgical and orthodontic treatment is often required to manage asymmetric cases. Advancement through a bilateral sagittal split osteotomy (BSSO) is generally performed to improve mandibular deviation. However, there is a risk of neurosensory sequelae to the inferior alveolar nerve3,4 and the BSSO is reportedly associated with a lack of stability if the mandibular procedure requires a significant advancement.5

In 1992, the first clinical report of mandibular distraction osteogenesis (DO) was published by McCarthy et al.6 The incidence of neurosensory disturbance is reported to be lower in DO compared with mandibular BSSO.7 Recently, DO has been performed to improve significant mandibular deviation in conditions such as hemifacial microsomia, juvenile chronic arthritis, and TMJ ankylosis.2,8,9 However, ideal occlusion cannot be achieved with DO alone because of difficulty in positioning the jaws in a spatially appropriate relationship. Subsequent orthognathic surgery may therefore be necessary to achieve satisfactory jaw position. DO and a BSSO...
have been shown to achieve a similar level of skeletal stability in cases that involve less than 7 mm of advancement. The results of skeletal stability, in cases that involve more than 10 mm of mandibular advancement, remain unclear.

The present case report describes a combination of surgical and orthodontic treatment of a 16-year-old female with a severe maxillary occlusal cant and a mandibular deviation caused by asymmetrical growth after a condylectomy to manage TMJ ankylosis. The case demonstrates that orthodontic treatment with accompanying DO and orthognathic surgery improved the patient’s facial asymmetry, which was maintained during the retention period.

**Diagnosis and aetiology**

The patient, a female aged nine years and six months, presented to the Osaka University Dental Hospital with the chief complaint of mandibular deviation. The patient suffered a facial injury at five years of age. Six months after the injury, she was unable to open her mouth and was diagnosed with right TMJ ankylosis. Six months later, a right side unilateral condylectomy was performed and at nine years and nine months, she was provided with a hybrid functional appliance to prevent further deterioration of a maxillary occlusal cant. At 16 years and four months, the functional appliance was discontinued.

Shortly thereafter, intraoral and extra-oral examinations were performed in preparation for a second phase of orthodontic treatment. The patient developed extreme facial deviation in addition to a convex profile and a retruded chin (Figures 1 and 2). She also developed an open bite and mild crowding in both arches (Figure 3). A posteroanterior cephalometric radiograph revealed that the mandible had deviated 15 mm to the right of the facial midline. In addition, there was a significant occlusal cant (Figure 4). A lateral cephalometric analysis indicated a skeletal Class II base relationship with an SNA of 76º, an SNB of 65º, producing an ANB of 11º (Table I). A high mandibular plane angle was noted with an SN-MP of 45º. The upper incisors were palatally inclined and the lower incisors were labially inclined. Computed tomography (CT) showed that the right condyle had separated between the condylar head and neck (Figure 6). It was determined that the patient required a combination of surgical and orthodontic treatment.

![Figure 1. Facial photographs. A: pretreatment; B: pre-surgery; C: post-treatment; D: post-retention, two years and six months after surgery.](image-url)
Figure 2. Facial photographs in full smile. A: pretreatment; B: pre-surgery; C: post-treatment; D: post-retention, two years and six months after surgery.

Figure 3. Oral photographs. A: pretreatment; B: pre-surgery; C: post-treatment, one year after surgery; D: post-retention, two years and six months after surgery.

**Treatment objectives**

The treatment objectives were: (1) to correct the maxillary occlusal cant and mandibular deviation; (2) to correct the skeletal Class II base relationship; (3) to co-ordinate the upper and lower dental arches; and (4) to achieve a Class I molar relationship and an ideal incisor relationship.

**Treatment alternatives**

It was considered that the treatment plan would require the alignment and co-ordination of the upper and lower dental arches coupled with the surgical correction of the maxillary occlusal cant, mandibular deviation, and skeletal Class II base relationship. (1) The first treatment option was to consider
two-stage surgery, the first stage of which aimed to intrude the maxilla in the left molar region and extrude in the right molar region by a Le Fort 1 osteotomy and to secondarily advance the right mandibular ramus with unilateral DO. A later second procedure was to rotate the mandible forward and upward with unilateral sagittal split osteotomy (USSO). (2) An alternative surgical consideration was to perform a Le Fort 1 maxillary osteotomy and a BSSO of the mandible.

The first treatment option was accepted because the BSSO was likely to involve a high level of risk due to the large amount of required mandibular advancement.
Treatment progress

At 17 years and one month, a transpalatal arch and a bihelix appliance were placed in the upper and lower arches to co-ordinate the dental arch widths. At 17 years and seven months, pre-adjusted edgewise appliances (0.022 x 0.028 inch slot) were placed in both arches and initial levelling was started using 0.014 inch nickel titanium wires. During the levelling and aligning phase, expansion of the lower arch was promoted by the bihelix appliance. At 19 years and seven months, pre-surgical orthodontic treatment was completed (Figure 3) and the surgical intrusive and extrusive correction of the maxilla was attempted by a Le Fort I osteotomy followed by the lengthening of the right mandibular ramus by unilateral DO. The distraction was started on postoperative day seven employing an elongation rate of 1.0 mm per day. The distraction was stopped after the ramus was elongated 23 mm (Figures 5 and 6). Three weeks after the completion of the DO, the left mandibular ramus was rotated forward and upward by a USSO. Miniscrews were placed in the mandibular buccal shelves during the USSO to prevent tooth extrusion by intermaxillary elastics that were to be used during the post-surgical orthodontic phase. Three months after surgery the distractor was removed, and after 12 months of post-surgical orthodontic treatment the patient was debonded and removable wrap-around retainers and fixed lingual bonded retainers were provided.

Treatment results

The patient's facial deviation and retruded chin significantly improved (Figures 1 and 2). The maxillary occlusal cant also greatly improved. The upper and lower dental midlines were centered with the facial midline. Ideal overjet and overbite were obtained. A cephalometric analysis showed that the patient had a skeletal Class I base relationship after orthognathic treatment (Table I). The lower incisor to mandibular
plane angle reduced by $13^\circ$ to $104^\circ$, suggesting lingual retraction of the lower labial segment by pre-surgical orthodontic treatment and expansion of the lower arch. Superimposed tracings of pre- and post-treatment lateral cephalometric radiographs indicated remarkable improvements in the facial profile and the anteroposterior position of the mandible (Figure 7). Furthermore, 3D volumetric superimposition indicated that the deviation of the mandible markedly improved (Figure 8).

At two years after the DO, skeletal stability was maintained. After one year of retention, the occlusion remained stable with a normal overjet and overbite (Figure 3). The improvements obtained in facial appearance and the dental arches during active treatment were preserved during the retention period, which satisfied the patient.

**Discussion**

Facial trauma is considered to be a major cause of TMJ ankylosis. TMJ mobilisation and/or condylectomy are often performed for patients with limited mandibular movement following TMJ ankylosis. However, condylectomy causes scarring in paediatric cases and growth on the affected side may be altered, and may lead to a mandibular deficiency asymmetry. Since the presented patient showed limited mouth opening due to the TMJ ankylosis after the facial injury, there was no alternative but to perform early condylectomy. Subsequently, a hybrid functional appliance was used to manage the unilateral deficiency in growth. It has been reported that patients affected by TMJ ankylosis who are treated with an activator show more favourable treatment results related to facial asymmetry and occlusal cant. Although the present patient wore this appliance for
seven years, the occlusal cant remained at the end of the first functional phase of treatment. Therefore, orthognathic surgery was considered to improve her occlusal cant and mandibular asymmetry during a second phase of therapy.

Specific surgical procedures, including BSSO, USSO, and DO, have been reported to improve severe facial asymmetry. A BSSO is limited in the degree of mandibular advancement that is possible if skeletal stability is to be maintained after surgery and USSO involves the risk of rotating the condyle on the non-surgical side. In addition, a BSSO is reported to be associated with a higher risk of neurosensory disturbance than DO. Therefore, DO seems to have advantages over the two alternative procedures, but achieving an optimal improvement of the occlusion with DO alone is complicated because of the difficulty in placing the mandible in a spatially appropriate position. Consequently, two-stage surgery was chosen, which included a Le Fort 1 maxillary osteotomy and a lengthening of the mandibular ramus with DO followed by a USSO. The major advantage of the two-stage surgery was the establishment of a desirable occlusion immediately after surgery.

After lengthening of the mandibular ramus in an inferior direction by DO, a lateral open bite on the elongated side is often a result. However, in the present case, a lateral open bite was observed on the opposite side because the mandibular body was chiefly increased in an anterior direction. Intermaxillary elastics or a functional appliance may be used to correct an open bite as a result of DO. Time is also required to correct an open bite and tooth extrusion was expected to occur in the meantime, which could have reduced occlusal stability. Therefore, miniscrews were placed to prevent the expected tooth extrusion by the use of intermaxillary elastics after the USSO-induced mandibular rotation.

Treatment timing is an important consideration in young patients with jaw deviation and DO is often performed to improve facial asymmetry during growth in patients with hemifacial microsomia. However, DO is associated with disadvantages that include occlusal change with continuing mandibular growth, possible relapse after the procedure, and the possible requirement of additional orthognathic surgery. In addition, there is no evidence of long-term stability of DO performed at an early time period. It was therefore decided to perform DO after the completion of mandibular growth to reduce the number of surgical procedures with the expectation of achieving optimal occlusal stability.

Figure 8. A superimposition of 3D CT images of the pretreatment: white; and post-treatment: blue. A: frontal view; B: inferior view.
A large amount of mandibular advancement and a high mandibular plane angle are considered to be risk factors related to DO instability.2,28 Recent studies have indicated that there is no statistical difference between DO and BSSO in post-surgical skeletal stability in cases involving less than 7 mm of mandibular advancement.2,29 However, large BSSO mandibular advancements have a higher likelihood of relapse.30 In the present case, despite the large amount of mandibular advancement and the high mandibular plane angle, relapse was not evident two years after DO. With regard to long-term stability, the results after DO procedures that involve more than 10 mm of mandibular advancement are unclear and require long-term review.

Conclusions
Orthodontic treatment incorporating distraction osteogenesis and orthognathic surgery improved a case presenting with a severe maxillary occlusal cant and mandibular deviation that had been caused by asymmetrical growth following condylectomy for TMJ ankylosis. Although more than 15 mm of unilateral mandibular lengthening was achieved, skeletal and occlusal stability was maintained after one year of retention, and two years after DO. It is considered that the combined surgical approach using DO and USSO is effective in cases of severe facial asymmetry.

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References
1. Munro IR, Chen YR, Park BY. Simultaneous total correction of temporomandibular ankylosis and facial asymmetry. Plast Reconstr Surg 1986;77:517-29.
2. Kaban LB, Bouchard C, Troulis MJ. A protocol for management of temporomandibular joint ankylosis in children. J Oral Maxillofac Surg 2009;67:1966-78.
3. Maclntosh RB. Experience with the sagittal osteotomy of the mandibular ramus: a 13-year review. J Maxillofac Surg 1981;9:151-65.
4. Bothur S, Blomqvist JE. Patient perception of neurosensory deficit after sagittal split osteotomy in the mandible. Plast Reconstr Surg 2003;111:373-7.
5. Will LA, West RA. Factors influencing the stability of the sagittal split osteotomy for mandibular advancement. J Oral Maxillofac Surg 1989;47:813-8.
6. McCarthy JG, Schreiber J, Karp N, Thorne CH, Grayson BH. Lengthening the human mandible by gradual distraction. Plast Reconstr Surg 1992;89:1-8.
7. Al-Moraisi EA, Ellis E 3rd. Bilateral sagittal split ramus osteotomy versus distraction osteogenesis for advancement of the retrognathic mandible. J Oral Maxillofac Surg 2015;73:1564-74.
8. Takashima M, Kitai N, Mori Y, Murakami S, Kreiborg S, Takada K. Mandibular distraction osteogenesis using an intraoral device and bite plate for a case of hemifacial microsomia. Cleft Palate Craniofac J 2003;40:437-45.
9. Singer SL, Southall PJ, Rosenberg I, Gillett D, Wülters M. Mandibular distraction osteogenesis and maxillary osteotomy in a class II division 1 patient with chronic juvenile arthritis. Angle Orthod 2006;76:341-8.
10. Kaban LB, Perrott DH, Fisher K. A protocol for management of temporomandibular joint ankylosis. J Oral Maxillofac Surg 1990:48:1145-51; discussion 1152.
11. Roychoudhury A, Parkash H, Trikha A. Functional restoration by gap arthroplasty in temporomandibular joint ankylosis: a report of 50 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1999;87:166-9.
12. Profit W, Fields H, Sarver D. Contemporary Orthodontics. 5th edn. St. Louis.: Elsevier Mosby, 2013;124.
13. Behnia H, Motamedi MH, Tehranchi A. Use of activator appliances in pediatric patients treated with costochondral grafts for temporomandibular joint ankylosis: analysis of 13 cases. J Oral Maxillofac Surg 1997;55:1408-14.
14. Altug-Atac AT, Grayson BH, McCarthy JG. Comparison of skeletal and soft-tissue changes following unilateral mandibular distraction osteogenesis. Plast Reconstr Surg 2008;121:1751-9.
15. Wohlwender I, Daake G, Weingart D, Brandstätter A, Kessler P, Lethaus B. Condylar resorption and functional outcome after unilateral sagittal split osteotomy. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;112:315-21.
16. Peacock ZS, Lee JS. Modification of the bilateral sagittal split osteotomy for the asymmetric mandible. J Oral Maxillofac Surg 2011;69:2437-41.
17. Schendel SA, Epker BN. Results after mandibular advancement surgery: an analysis of 87 cases. J Oral Surg 1980;38:265-82.
18. Cutbirth M, Van Sickels JE, Thrash WJ. Condylar resorption after bicortical screw fixation of mandibular advancement. J Oral Maxillofac Surg 1998;56:178-82.
19. Borstlap WA, Stoelinga PJ, Hoppenreijs TJ, van’t Hof MA. Stabilisation of sagittal split advancement osteotomies with miniplates: a prospective, multicentre study with two-year follow-up. Part II. Radiographic parameters. Int J Oral Maxillofac Surg 2004;33:535-42.
20. Schreuder WH, Jansma J, Bierman MW, Vissink A. Distraction osteogenesis versus bilateral sagittal split osteotomy for advancement of the retrognathic mandible: a review of the literature. Int J Oral Maxillofac Surg 2007;36:103-10.
21. Tehranchi A, Behnia H. Treatment of mandibular asymmetry by distraction osteogenesis and orthodontics: a report of four cases. Angle Orthod 2000;70:165-74.
22. Amm EW. Three-year follow-up of a patient with hemifacial microsomia treated with distraction osteogenesis, temporary anchorage devices, and orthodontics. Am J Orthod Dentofacial Orthop 2012;142:115-28.
23. McCarthy JG, Katzen JT, Hopper R, Grayson BH. The first decade of mandibular distraction: lessons we have learned. Plast Reconstr Surg 2007;111:373-7.
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24. Molina F. Mandibular distraction osteogenesis: a clinical experience of the last 17 years. J Craniofac Surg 2009;20 Suppl 2:1794-800.
25. Meazzini MC, Mazzoleni F, Bozzetti A, Brusati R. Comparison of mandibular vertical growth in hemifacial microsomia patients treated with early distraction or not treated: follow up till the completion of growth. J Craniomaxillofac Surg 2012;40:105-11.
26. Nagy K, Kuijpers-Jagtman AM, Mommaerts MY. No evidence for long-term effectiveness of early osteodistraction in hemifacial microsomia. Plast Reconstr Surg 2009;124:2061-71.
27. Molina F, Ortiz Monasterio F. Mandibular elongation and remodeling by distraction: a farewell to major osteotomies. Plast Reconsr Surg 1995;96:825-40.
28. van Strijen PJ, Breuning KH, Becking AG, Tuinzing DB. Stability after distraction osteogenesis to lengthen the mandible: results in 50 patients. J Oral Maxillofac Surg 2004;62:304-7.
29. Baas EM, Bierenbroodspot F, de Lange J. Skeletal stability after bilateral sagittal split osteotomy or distraction osteogenesis of the mandible: a randomized clinical trial. Int J Oral Maxillofac Surg 2015;44:615-20.
30. Schendel SA, Epker BN. Results after mandibular advancement surgery: an analysis of 87 cases. J Oral Surg 1980;38:265-82.