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British Journal of Oral and Maxillofacial Surgery 57 (2019) 135–139

BRITISH Journal of Oral and Maxillofacial Surgery

www.bjoms.com

Simultaneous arthroplasty and distraction osteogenesis for the treatment of ankylosis of the temporomandibular joint and secondary mandibular deformities in children

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Accepted 26 November 2018

Available online 29 December 2018

Abstract

The purpose of this study was to explore the use of simultaneous arthroplasty and distraction osteogenesis in the treatment of children with ankylosis of the temporomandibular joint (TMJ) and secondary mandibular deformities. Between January 2012 and December 2016, 17 children (7 boys and 10 girls, mean (range) age 7 (4–12) years) were treated. Preoperatively, the mean (range) maximal incisal opening was 1.4 (0–5) mm. Distraction osteogenesis was used to elongate the mandibular body or ramus, or both, after the release of ankylosis. Distraction began after five to seven days at a rate of 0.5 mm twice daily, and the distractor was removed three to five months after the completion of distraction. The mean (range) follow-up time after removal was 29.6 (16–45) months, and the distance of distraction was 14.4 (10–18) mm. After treatment, all patients had satisfactory outcomes, a good facial profile, alignment of the midline lower incisor, and a level occlusal plane. The mean (range) maximum incisal opening reached 35.7 (31–41) mm. Bone formation across the distraction gap was good. The mean minimum axial area of the airway increased from 61.4 mm to 96.4 mm ($p < 0.01$). No patients had a recurrence of ankylosis during follow up. Our results suggest that simultaneous arthroplasty and distraction osteogenesis is feasible in this group.

Keywords: distraction osteogenesis; temporomandibular joint; ankylosis; mandibular deformities; children

Introduction

Ankylosis of the temporomandibular joint (TMJ) is often the result of trauma or infection, and severely affects function.[1] Currently, it is common in developing countries. When it occurs during early childhood, it alters mandibular growth and leads to progressive facial deformities that include micrognathia and occlusal discrepancy;[2,3] it can also cause severe psychosocial problems.[4] The treatment of patients who also have secondary dentofacial deformities is challenging.

Distraction osteogenesis is a technique that involves use of a mechanical device to move two vascularised bony surfaces gradually apart, causing new bone to form in the gap between the two ends of cut bone.[5,6] A distinctive feature is that the regeneration is accompanied by simultaneous

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https://doi.org/10.1016/j.bjoms.2018.11.016

expansion of the functional soft tissue matrix (histogenesis).[7] Many authors have described the use of distraction osteogenesis for the treatment of patients with ankylosis of the TMJ and secondary mandibular deformities, including reconstruction of the mandibular condyle, and lengthening of the mandibular ramus and body.[1]

In this study, we present and discuss our experience of simultaneous arthroplasty and distraction osteogenesis for the treatment of ankylosis of the TMJ and secondary mandibular deformities in children.

Patients and methods

Patients

Between January 2012 and December 2016, we studied 17 children (seven boys and 10 girls, mean (range) age 7 (4–12) years) with ankylosis of the TMJ and secondary mandibular deformities. Ankylosis was bilateral in four and unilateral in 13. The mean (range) preoperative maximal incisal opening was 1.4 (0–5) mm. All patients had simultaneous arthroplasty and distraction osteogenesis at the Orthognathic and TMJ Surgery Centre, West China Hospital of Stomatology, Sichuan University. The study protocol was approved by the hospital's review board, and guardians of all the participants signed an informed consent agreement.

Patients with unilateral or bilateral ankylosis of the TMJ and secondary mandibular deformities who were under 15 years of age, were included. Patients and guardians gave their consent to participate, and no patient had surgical or anaesthetic contraindications.

Those without secondary mandibular deformities, or with surgical or anaesthetic contraindications (such as severe hypertension, or diseases of the heart, liver, or kidney), and whose parents or guardians did not give their consent, were excluded.

Operation

We removed the ankylotic bone through a preauricular incision to create a gap 1.0–1.5 cm wide (Fig. 1A), and used a superficial temporalis flap to cover the surface of the articular fossa. The residual dead space between the ramus and the glenoid fossa was then filled with a pedicled buccal fat pad. Patients with a passive maximum incisal opening of less than 30 mm after arthroplasty had coronoidectomy.[8] After release of the ankylosis, we created a transport disc at the ramus with an L-shaped or inverted L-shaped osteotomy through a submandibular incision (Fig. 1B). To ensure blood supply to the disc, we did not disturb the medial periosteum and muscle attachments. A distractor (Ningbo Cibei Medical Devices Co Ltd) was fixed with screws, and the distraction vector calculated with the help of virtual surgical planning. Careful planning was necessary to avoid damage to the inferior alveolar nerve.

Distraction and consolidation period

After a latency period of five to seven days, distraction was done at a rate of 0.5 mm twice a day. All patients initially had a liquid diet, and began mouth-opening training seven days after the operation to avoid a recurrence of ankylosis. We calculated the distance of distraction by clinical observation of the facial profile, alignment of the midline lower incisor, and use of radiographs including spiral computed tomograms (Philips Brilliance 16-slice scanner). Patients had monthly panoramic radiographs to assess bony growth at the distraction gap, and after three to five months of consolidation, the distraction devices were removed.

Clinical evaluation

We based the assessment of outcome on oral function, radiography, and medical photography, and used medical photographs and panoramic radiographs, as well as computed

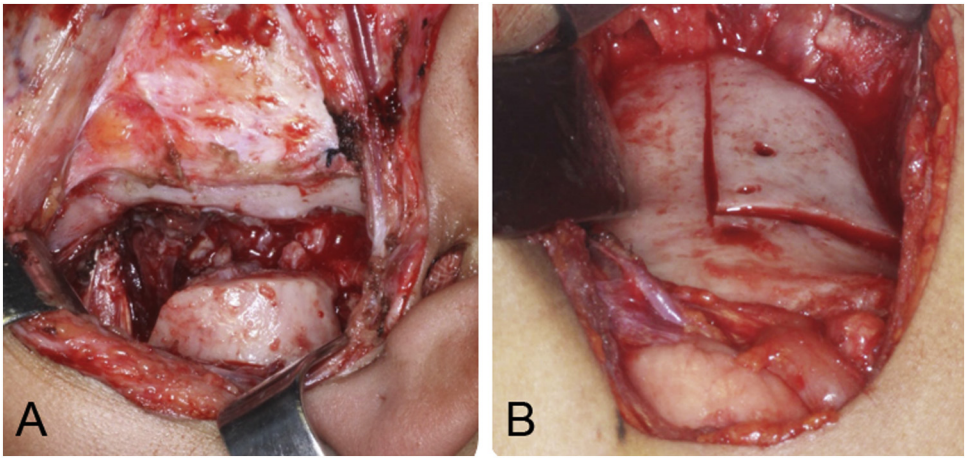

Fig. 1. Operations. A: gap arthroplasty; B: "L"-shaped osteotomy for transport distraction osteogenesis.

Table 1
Length of distraction, follow-up time, and changes in mouth opening and posterior airway. Data were obtained preoperatively and six months after removal of the distractor.

Case No.	Maximal incisal opening (mm)		Distraction (mm)	Follow-up time (months)	Minimum axial area (mm^2)	
	Before	After			Before	After
1	0	35	15	33	79	101
2	0	32	18(L), 16(R)	24	80	109
3	2	37	15	45	78	95
4	0	41	13(L), 14(R)	41	24	50
5	0	36	14	19	37	83
6	3	33	13	34	62	92
7	0	41	14	16	83	133
8	3	39	13	24	90	128
9	5	37	15	38	46	90
10	3	31	17	25	53	92
11	0	34	17	23	88	109
12	0	39	16	33	46	97
13	0	35	14(L), 15(R)	36	38	100
14	1	32	10	42	29	55
15	0	33	16(L), 16(R)	30	86	110
16	2	37	13	18	81	95
17	4	35	11	22	43	99
Mean	1.4	35.7	14.4	29.6	61.4	96.4

L: left; R: right.

tomograms (six months after removal of the distractor), to assess changes in the airway. Patients had monthly panoramic radiographs after operation to assess the formation of bone, and we recorded maximum incisal opening before operation and at the end of follow up. We used Dolphin imaging software (Patterson Dental) to reconstruct the upper airway, and calculated the minimum axial area of the airway. There was no formal assessment of obstructive sleep apnoea before or after the procedure.

Statistical analysis

Statistical analysis was done with the help of SPSS version 10.0 (SPSS Inc). The minimum axial area of the airway before and after treatment was compared using the paired *t* test. Probabilities of less than 0.05 were considered significant.

Results

Outcomes

All patients tolerated the operation well and there were no complications during their stay in hospital. However, three patients had a local wound infection during the consolidation period that was controlled with irrigation and dressing. The mean (range) follow-up time was 29.6 (16–45) months, and there were no signs of a recurrence of ankylosis. The mean (range) maximum incisal opening before operation was 1.4 (0–5) mm. At the end of follow up it was 35.7 (31–41) mm. The mean (range) distance of distraction was 14.4 (10–18) mm. The mean minimum axial area of the upper airway increased from 61.4 mm to 96.4 mm ($p < 0.01$) (Table 1). Consecutive radiographs showed good formation of bone even when there was local infection. All patients had satisfactory outcomes and there were considerable improvements in appearance and mouth opening. During follow up, there was no relapse of the distracted mandible.

Case 1

A 12-year-old girl had simultaneous arthroplasty and distraction osteogenesis to treat ankylosis of the left TMJ and severe facial asymmetry (Fig. 2A). We calculated the distraction vector using virtual planning (Supplemental Fig. 1), and removed the distractor four months after the completion of distraction. Her facial asymmetry was corrected and she had a normal facial profile (Fig. 2B). Maximum incisal opening had increased from 0 to 35 mm (Supplemental Fig. 2), and the minimum axial area of the upper airway from 79 mm to 101 mm (Supplemental Fig. 3). Computed tomographic (CT) reconstruction showed considerable increases in the length and height of the affected mandible (Supplemental Fig. 4). Immediately after the completion of distraction she had an ipsilateral lateral open bite. This subsequently closed with compensatory maxillary dentoalveolar growth that levelled the occlusal cant (Supplemental Fig. 5).

Case 2

A 9-year-old girl had simultaneous bilateral gap arthroplasty and distraction osteogenesis to treat bilateral ankylosis of the TMJ, severe mandibular retrusion, and obstructive sleep apnoea. The distractor was removed four months after the completion of distraction. The mandibular retrusion was corrected, but not sufficiently. The maximum incisal opening

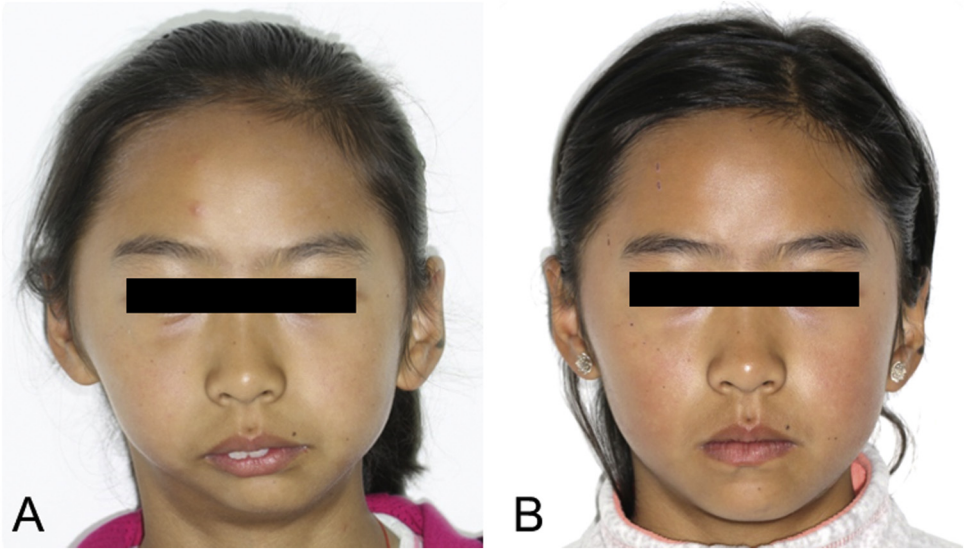

Fig. 2. Changes in facial profile after arthroplasty and transport distraction osteogenesis. A: before treatment; B: 6 months after the distractor had been removed.

had increased from 0 to 32 mm, and the minimum axial area of the upper airway from 80 to 109 mm. Obstructive sleep apnoea had almost disappeared (Supplemental Fig. 6).

Discussion

Ankylosis of the TMJ that develops in childhood leads to mandibular deformity and an impairment in growth.[4] Recently, distraction osteogenesis has become widely accepted as a treatment for mandibular hypoplasia and asymmetry, as well as obstructive sleep apnoea,[9–11] and it has a great potential for treating difficult cases, particularly when other methods have failed.[12]

Options for the reconstruction of the mandibular condyle, which can be difficult because of the anatomy and physiology of the TMJ,[13] include autogenous bone grafts and alloplastic materials. The harvest of bone grafts, however, can result in donor site morbidity. Grafts can be an inappropriate size or design, or can fail, and resorption can lead to a recurrence of the skeletal deformity.[14,15] The cost of alloplastic materials is still a great concern for patients in developing countries, and artificial materials that have no potential for growth are usually used in adults and not in children.

Transport disc distraction osteogenesis can effectively reconstruct the mandibular condyle,[13,16] and enable reconstruction of a new TMJ and correction of a deficiency in ramus height.[3] Compared with a free bone graft, it has a lower risk of bony necrosis and resorption, because the living transport segment can preserve a continuous blood supply.[15] There was no further ankylosis during follow up in all our patients, and recurrence was prevented by adequate gap arthroplasty, function (aided by physiotherapy), and filling of the gap with temporalis fascia and a pedicled buccal fat pad.

Dentomaxillofacial deformities secondary to ankylosis of the TMJ in children severely affect their mental health and should be treated as early as possible. However, indications and considerations for correction differ considerably between growing and skeletally-mature patients.[4] Orthognathic surgery, genioplasty, and jaw contouring, are common treatments for the correction of facial deformities in adults, but cannot be used in growing childen. Autologous reconstruction (costochondral grafts, free fibular flaps) and total alloplastic replacement joints can also correct mandibular underdevelopment, but the forces from the adjacent muscles, particularly contraction of the suprahyoid depressor muscles, increase the difficulty of the operation and the risk of relapse.[3] Distraction osteogenesis can lengthen the mandible and simultaneously elongate both the soft tissues and bone during distraction, and result in more stable, functional, and aesthetic outcomes.[17] When done in conjunction with gap arthroplasty, it has successfully treated ankylosis of the TMJ in children.

Ankylosis disturbs mandibular growth and facial skeletal development. Costochondral grafts have been used to enable growth, but have been unpredictable.[18,19] The potential of the mandible to grow must be taken into account when we choose how to correct deformities in children. Distraction osteogenesis is a simpler, safer procedure that causes less morbidity than traditional orthognathic surgery, but questions regarding growth have not been clearly explained.[20–22] Opinions therefore may differ about the amount of distraction needed to obtain stable results, as we cannot predict growth patterns. Should we distract the mandible by the exact amount needed, or by more to account for future development? We used the occlusal level and the tooth in the midline of the mandible to calculate the exact amount needed. The deformities were successfully corrected, but with ongoing growth

of the undamaged tissues and failure of the reconstructed tissues to grow, further treatment may be necessary.

Distraction osteogenesis has some disadvantages, one of which is the need for a second operation to remove the distractor. Another is the length of treatment.[23] Other complications are mainly associated with technical failures such as "pin pullout", exposure of hardware, unsatisfactory occlusion, and asymmetry as a result of distraction along an inappropriate vector.[24,25] Careful preoperative design and intraoperative technique is key to reducing or avoiding the incidence of these complications. Better-designed distractors and the promotion of bony formation may encourage wider use of the approach.

In conclusion, simultaneous arthroplasty and distraction osteogenesis is an effective treatment for ankylosis of the TMJ and secondary mandibular deformities in children. The long-term influence of distraction on mandibular growth, however, needs further investigation.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patients' permission

The study protocol was approved by the West China Hospital of Stomatology Institutional Review Board. Patients' permission was obtained.

Acknowledgements

This study was supported by a grant from the National Natural Science Foundation of China (No. 81271170).

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.bjoms.2018.11.016.

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