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Case report

Release of syngnathia by anticlockwise rotation and mandibular advancement using bilateral alloplastic temporomandibular joint prostheses: a new approach

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Abstract

We describe a new approach to the planning of treatment and subsequent operation on a patient with syngnathia and severe mandibular retrognathism. To facilitate a large mandibular advancement we applied alloplastic temporomandibular joint (TMJ) prostheses to the coronoid processes after anticlockwise rotation of the mandible. To the best of our knowledge this is the first documented case of its kind.

Keywords: Syngnathia; Pierre–Robin syndrome; TMJ prostheses; coronoid; counter-clockwise rotation

Introduction

Congenital or acquired micrognathia poses many challenges. Apart from the obvious aesthetic concerns, most patients present with severely compromised oropharyngeal airways, and the associated increased risk of obstruction.[1]

Most cases of congenital micrognathia may be attributed to any one of the following syndromes: Pierre–Robin syndrome, hemifacial microsomia, Goldenhar syndrome, Nager syndrome, Treacher Collins syndrome, and condylar hypoplasia. Syngnathia may be caused by injury (in utero or during infancy), and septic arthritis of the temporomandibular joint (TMJ) may cause micrognathia; both of which are secondary to ankylosis of the joint.

Syngnathia is the congenital fusion of the maxilla to the mandible through fibrous bands (synechiae) or a bony fusion (synostosis).[2] Synostosis can develop between the maxillary tuberosity and mandibular coronoid process; the maxillary and mandibular alveoli; the mandible and zygomatic complex; and along the pterygomandibular raphe. Managing syngnathia is challenging because early intervention is required, but often this presents the greatest risk for reattachment of the bony junctions. Our procedure was designed to release the syngnathia and address some complications of Pierre–Robin syndrome.

Case report

A 19-year-old man was diagnosed with Pierre–Robin syndrome, (specifically Siebold–Robin sequence)[2] that was complicated by the presence of congenital syngnathia. Ossification of the pterygomandibular raphe had presented as mandibular retrognathia with micrognathia (Fig. 1). He had had a tracheostomy in place since infancy, and had had two previous operations to release the syngnathia with attempted mandibular distraction osteogenesis (2000 and 2012). In 2016 he presented again, and requested better facial aesthetics, the ability to eat normally, and to have his tracheostomy tube removed.

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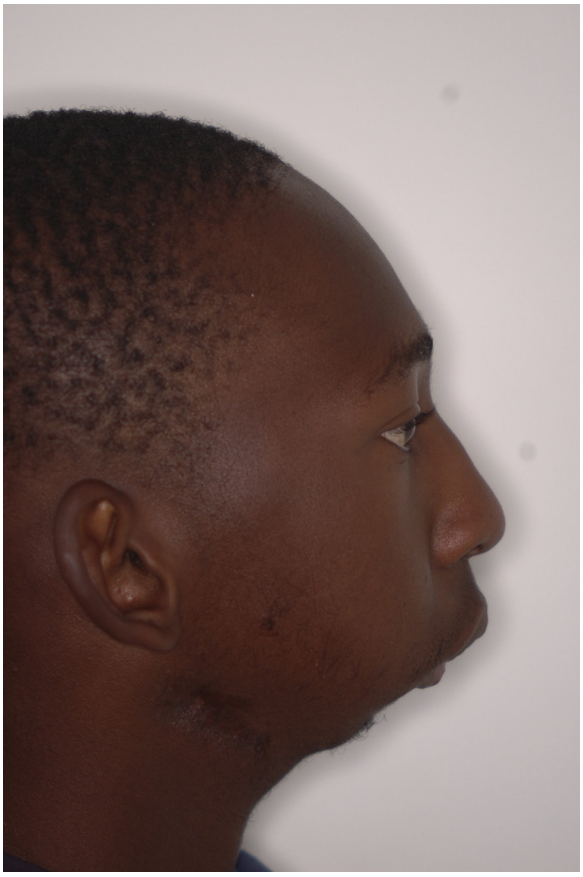

Fig. 1. Preoperative photograph showing mandibular retrognathia with micrognathia.

Operative planning consisted of computed tomography (CT) and a 3-dimensional stereolithographic model. The operation comprised the release of the syngnathia (secondary to a left corpus mandibulotomy), bilateral condylectomies, partial coronoidectomies, and selective stripping of the masticatory musculature. This was followed by an anticlockwise mandibular rotation with advancement, and the placement of bilateral TMJ prostheses on the coronoid processes (Fig. 2). Dental implants would later be considered for the anterior mandible.

Bilateral preauricular and submandibular approaches were favoured for the bony operations. We stripped off the temporalis and lateral pterygoids, and the pterygomasseteric slings, the latter of which were later reattached. We rotated the mandible anticlockwise and stabilised it using maxillomandibular fixation screws and wire. A pair of alloplastic TMJ stock prostheses (Zimmer Biomet) were placed on both coronoid processes to create a new functional, repositioned joint. We repaired the mandibulotomy using a 2.4 mm plate and screws (DePuy Synthes), placed a nasogastric feeding tube for two weeks, and gave him antibiotics and analgesia for a week. Twenty sessions of hyperbaric oxygen therapy were given to improve recovery. Two weeks after operation, we removed the tracheostomy and his speech was much

Fig. 2. Three-dimensional stereolithographic model showing the placement of bilateral temporomandibular joint prostheses on the coronoid processes.

improved. Four weeks postoperatively he was able to eat a soft solid diet and was discharged from hospital.

Follow-up was biweekly for two months. Since then he has been closely followed up for 18 months and recalled every 12 weeks. Healing has been unremarkable, with no sepsis or neurological deficit. He has maintained good mouth opening, and has been able to eat a normal solid diet. The biggest benefits of the procedure have been a marked improvement in facial aesthetics, and the ability to maintain his airway unassisted.

Discussion

Three primary objectives were addressed: to create mouth opening and mandibular mobility; to increase the diameter of the oropharyngeal airway and; to improve the facial soft tissue profile (Fig. 3). Overall mouth opening improved from 0 mm to 19 mm postoperatively, and there has been full preservation of sensory and motor innervation. Mandibular advancement increased the diameter of the airway from 3.1 mm to 8.6 mm, and the improvement in the patency of the airway allowed for the removal of tracheostomy tube after 18 years. Our experience agreed with the findings of Coleta et al, who found an improvement in the dimensions of the lumen of the airway after anticlockwise mandibular rotation.[4]

The use of alloplastic TMJ prostheses allows for larger than usual anticlockwise rotations of the mandible as there are no restrictions caused by muscular or capsular attachments. This facilitated a large advancement without the need for distraction osteogenesis or sagittal osteotomies of the mandible.[5]

We would have preferred an advancement to a class I relation but, because of limited resources, we had to rely on stock prostheses placed on the coronoid processes, as the bodies of the mandibular posterior rami were absent. Previous operations meant that the muscles and soft tissues were fibrosed and unyielding, which only allowed for advancement to a class II skeletal relation.

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Fig. 3. Postoperative photograph showing advancement to a class II skeletal relation.

Congenital syngnathia is rare, with synechia being more common than synostosis.[2,6,7] In this case, it is plausible to assume that the Pierre–Robin syndrome was a consequence of the congenital syngnathia. The bony attachment of the mandible and its consequent immobility would probably have hindered mandibular growth, and resulted in micrognathia, retrognathia, glossoptosis, and microglossia.

Using the coronoid processes for the placement of alloplastic TMJ prostheses allows for the correction of mandibular retrognathia, and the anticlockwise rotation improves the angle of the mandibular plane and that of the chin and throat. In the future this technique will benefit both surgeons and patients by reducing the number of operations necessary for the treatment of syngnathia and mandubular retrognathism.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patient's permission

No ethics approval required. Written consent was obtained from the patient.

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