

Pterygomandibular ankylosis: a report on two cases

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ABSTRACT

Pseudo-ankylosis of the temporo-mandibular joint (TMJ) refers to a persistent restricted mandibular hypomobility resulting from a pathologic condition outside the TMJ. Ankyloses may be acquired or congenital. Most congenital ankyloses are extracapsular (pseudo- ankyloses), while most of the acquired ankyloses (true ankyloses) are intracapsular. An extremely rare form of extracapsular ankylosis is bony pterygomandibular fusion, few cases having been reported in the literature. To our knowledge, a case with two separate ipsilateral extracapsular ankylosis sites together with bilateral intracapsular ankylosis has never been reported. This paper considers the case of a 24 year-old patient who had been unable to open his mouth for six years. There was neither a history of trauma nor one of recurrent infection. The patient presented with a slight facial asymmetry with deviation of the mandible towards the right. CT scans confirmed the diagnosis of a complex pseudoankylosis, characterised by unilateral bony fusions between the lateral pterygoid plate and the medial aspect of the ramus, coronoid process and zygoma together with bilateral intracapsular ankyloses. The patient underwent surgery for ostectomy of the pterygomandibular fusion and a bilateral coronoidectomy. Comments on a paediatric case with bilateral bony pterygomandibular fusions are also included.

INTRODUCTION

Pseudoankylosis of the temporo-mandibular joint (TMJ) refers to a persistent restricted mandibular hypomobility

ACRONYM

TMJ: temporo-mandibular joint

resulting from a pathologic condition outside the TMJ.¹ Kazanjian classified TMJ ankylosis into intracapsular (true) ankylosis and extracapsular (false/pseudo-ankylosis).² Ankyloses may be acquired or congenital. According to Allori *et al* 64% of the congenital ankyloses are extracapsular, while most of the acquired (true) TMJ ankyloses (85.7%)³ are intracapsular. A minority of extracapsular ankyloses may be myogenic, osteogenic, neurogenic or psychiatric.¹ The most common causes of acquired pseudoankylosis are trauma, radiation and infections.⁴ The congenital forms of pseudo-ankylosis include synechia and syngnathia.^{5,6} Various forms of extracapsular ankylosis have been described in literature including coronoid to base of the skull,⁷ syphilitic myositis involving masseter and temporalis,⁸ osteoma of the coronoid process, scar formation within the muscles of mastication and unilateral coronoid hyperplasia.¹ The most common form is coronoid-zygomatic ankylosis, which usually follows fracture of the zygoma.⁷ An extremely rare form of extracapsular ankylosis is bony pterygomandibular fusion, few cases having appeared in the literature.^{1,6,9,10} A bilateral form of pterygomandibular fusion has also been reported.⁷ Accurate diagnosis of pterygomandibular ankylosis requires a computed tomographic (CT) scan,^{5-7,8,9} while stereolithographic models add value in planning for surgery.⁵

To our knowledge, a case with two separate ipsilateral extracapsular ankylosis sites together with bilateral intracapsular ankylosis has never been reported in the literature.

The main purpose of this paper is to report on a case with a complex pseudoankylosis, characterised by unilateral bony fusions between the lateral pterygoid plate and the medial aspect of the ramus, coronoid process and zygoma together with bilateral intracapsular ankyloses. Observations on a paediatric case with bilateral bony pterygomandibular fusions are also included.

CASE REPORTS

Case 1

A five year old girl presented with inability to open the mouth. She had been delivered by caesarean section as a

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result of foetal distress. The following clinical features were noted at birth: hypoplastic mandible, limitation of mouth opening and low slung ears. Feeding difficulties were reported and needed to be supplemented with nasogastric feeding.



Figure 1: Restricted mouth opening of patient.

The patient was discharged from the hospital seven months after birth. At age of eleven months a tracheostomy was performed due to poor oxygen saturation. Clinical examination revealed a severely reduced lower third of the face with a hypoplastic mandible. The low slung ears were seen to be notched. No facial asymmetry, however, was present. Poor oral hygiene characterised by rampant caries (Figure 1) was noted.

Computed tomography showed normal joints on both sides, but bony fusion between the medial pterygoid plate and the medial aspect of the ramus of the mandible was noted (Figure 2).

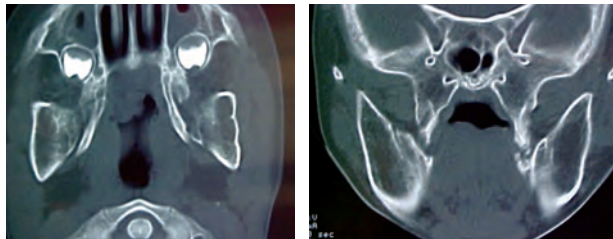


Figure 2: CT scans showing bony fusion between the pterygoid plate of the sphenoid bone and the medial aspect of the ramus of the mandible.



Figure 3: Mouth opening a year after the operation.

Poor compliance resulted in a delay of a year after the initial examination before surgical intervention could be undertaken to release of the pseudo-ankylosis. An incision was made over the oblique ridge of the mandible and subperiosteal stripping carried out on the lingual aspect of the ramus. A bony ridge was identified along the inferior aspect of the mandible, which was sectioned and small spicules of bone removed. The procedure

was carried out on both sides. Bilateral coronoidectomy was performed and an inter-incisal distance of 30mm was achieved postoperatively. Tracheostomy was removed after the procedure as the patient could maintain her airway. Mouth-opening exercises were begun 24 hours after the procedure and continued under supervision for a month. Thereafter the patient relocated to her rural home, returning a year later with severe restriction of mouth opening (Figure 3). Despite the fact that the patient was able to achieve a total mouth opening of only 15-20mm, her parents were not keen on a follow-up operation.

Case 2

A 24 year old male presented with a six year history of inability to open his mouth. There was neither a history of trauma nor one of recurrent infections. His general medical history was unremarkable. The patient presented with slight facial asymmetry characterized by deviation of the mandible towards the right and flattening of the left side of the face in the mandibular region. The maximal inter-incisal distance was zero. A panoramic radiograph revealed a pronounced antegonial notch, elongated coronoid process and shortening of the ramus height on the right side. The right lower wisdom tooth was vertically impacted.

CT scans demonstrated bony fusion between the left lateral pterygoid plate and the medial aspect of the mandible as well as fusion between the left coronoid process and the zygomatic bone. The joint space could be clearly appreciated on CT scans. Osteophytes in the right joint space could be seen as well as fusion of the lateral aspect of the left condylar head to the mandibular fossa. The three CT scans shown in Figure 5 clearly support the diagnosis of a right bony pterygomandibular ankylosis, a bony zygomatico-coronoid ankylosis and bilateral ankylosis of the TMJ.

The patient was admitted to theatre for ostectomy of the pterygomandibular fusion and a bilateral coronoidectomy. The zygomatic arch was removed on the right side to improve access and to facilitate an ostectomy of the zygomatico-coronoid ankylotic mass. The arch was replaced and fixed with plates and screws at the end of the procedure. Significant mouth opening was still not achieved, and there-

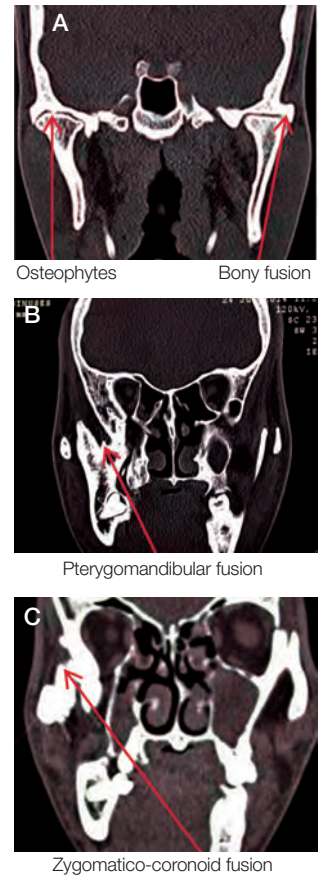


Figure 5: CT scans showing the site of ankylosis: **A** reveals flattening of the condylar head with bony fusion on the lateral aspect of the left condyle; **B** shows bony pterygomandibular fusion and a lingually inclined impacted 48; **C** illustrates zygomatico-coronoid fusion.

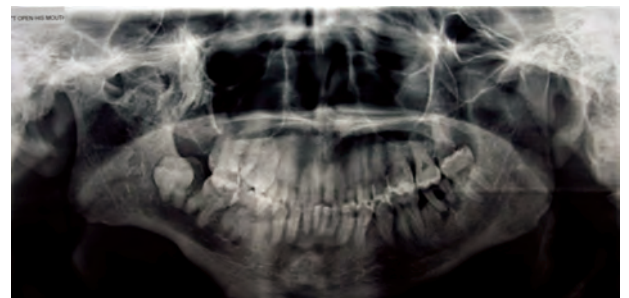


Figure 4: Panoramic radiograph of the patient.

fore exploration of the joints was performed. Exposure of the joints showed bony hyperplasia on the lateral aspect of both joints. Bilateral gap arthroplasty was then performed and arch bars placed on both jaws, enabling a mouth opening of 40mm. The patient slept well but was unable to close his mouth. Heavy elastics were then used to facilitate closure and to prevent anterior open bite. The patient was placed on a physiotherapy protocol. Three months after the procedure the mouth opening was 25mm but thereafter the patient was lost to follow up.

DISCUSSION

Few cases of pterygomandibular fusions have been reported in the literature,^{1,6,7,9,10} but these authors preferred to use the term “complex pseudo-ankylosis” to describe this condition as it involves two extracapsular bony fusions and a bilateral intracapsular component.

The main causes of acquired extracapsular ankyloses that have been reported in the literature are trauma, infections, irradiation and tumours,⁴ although a study by Allori *et al* in 2010 suggests that most extracapsular ankyloses are congenital in origin.³ Super and Cotton (1978) also suspected a congenital syndrome in their case of pterygomandibular fusion.⁹

Although the aetiology of the second case is idiopathic, the slight deviation of the mandible suggests that ankylosis occurred before the final growth spurt. Elongation of the coronoid process, followed by fusion between the coronoid process and the zygoma, and the intracapsular component would have resulted in limitation of mouth opening.

Although a CT scan is the gold standard for diagnosis of pterygomandibular bony fusions,¹ MRI is superior for fibrous pseudo-ankylosis.^{1,6} In the case of this report, plain x-rays were quite useful in identifying the area and in discerning certain pathological changes. Slice B of the CT scan shows a thick bony pterygomandibular fusion, while slice A shows degenerative changes and early bony fusion on the lateral aspect of the ipsilateral joint.

Simple pseudo-ankylosis refers to the ankylosis that is outside the joint without any radiographic changes on the articular surfaces of the TMJ. Complex pseudo-ankylosis on the other hand refers to a simple extracapsular ankylosis with secondary intracapsular degenerative changes and/or ankyloses.^{1,2} Based on our observations, however, we propose that pseudo-ankylosis of the TMJ be regarded as both simple and complex.

During surgery, discs were found fused to the mandibular fossae. We believe this phenomenon is a secondary response to lack of movement, which is followed by fibrosis and atrophy of the muscles of mastication.¹¹ Hence, exploration of the joints is mandatory when an extracapsular ankylosis is released.

The challenge faced postoperatively was that the patient could not close his mouth a day after the procedure, and

needed to push the lower jaw upward with his hand. This is believed to have been the result of bilateral coronoidectomies in a patient with atrophic and fibrotic muscles of mastication with loss of neural activity.¹¹ The poorly functional muscles of mastication were too weak to close the mouth while the mylohyoid, digastric and suprahyoid muscles together with gravity could depress the lower jaw after release of ankylosis. The problem was resolved by using arch bars and heavy elastics.

Outcomes in the management of patients with pterygomandibular fusion have not, on the whole, been very successful.^{8,10} Our patient could achieve a mouth opening of only 25mm three months after surgery. A structured post-operative physiotherapy regimen to repair and maintain adequate mandibular movement is essential for a successful outcome.

Conflict of interest: None declared.

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