

Unilateral bony ankylosis of the temporomandibular joint in a case of ankylosing spondylitis

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Abstract

Background Ankylosing spondylitis (AS) is a chronic inflammatory disease with multiple articular and para-articular involvement that has a predilection for the axial skeleton. In spite of its high prevalence, ankylosis secondary to AS is a rare condition.

Case report A 31-year-old male diagnosed with AS was referred for computed tomography (CT) of the temporomandibular joint (TMJ) due to severe mouth opening limitation. The patient

had a 16-year medical history of AS and sought assistance due to TMJ pain and incapacity to open his mouth.

Results Previous bony scintigraphy revealed involvement of the spine, sacroiliac joints, right knee, and left TMJ. Magnetic resonance imaging revealed erosion of the left condyle and posterior slope of the articular eminence, and a mass of heterogeneous signal intensity between these structures. The left condyle also presented sclerosis/edema of the bone marrow and the disk could not be identified. Sagittal and coronal CT images showed moderate alterations of the TMJ on the right side. On the left side, the images displayed markedly eroded condyle and mandibular fossa, and a bony mass resulting in ankylosis of the osseous components of the joint.

Conclusion TMJ ankylosis in AS patients is rare and very few reports have presented imaging features of the condition through advanced diagnostic techniques.

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Keywords Ankylosing spondylitis · Temporomandibular joint · Tomography · X-ray computed · Scintigraphy · Magnetic resonance imaging

Background

Ankylosing spondylitis (AS) is a chronic inflammatory disease with multiple articular and para-articular involvement that has a predilection for the axial skeleton [1, 2]. The clinical signs vary from mild mobility limitation to total ankylosis and can be compounded by extra-articular manifestations. Although the initial clinical signs are pain and discomfort, synovial changes progressively involve all the axial joints, including the temporomandibular joint (TMJ) [3].

TMJ shares the tendency of involvement by AS as with other fibrocartilagenous structures such as the symphysis pubis and the intervertebral disks [Chow 1997, [4]]. Involvement of the TMJ appears to give rise to few serious symptoms until pain, stiffness, and gross restriction of jaw movement have occurred. Flattening of the condyles and erosion of the cortical outline are the most common findings [Polites 1087].

Computed tomography (CT) and magnetic resonance imaging (MRI) images show temporal flattening, abnormal condylar shape, erosions, sclerosis, disk alterations, osteophytes, and even total ankylosis of the joint (Koidis 2009)

In spite of its high prevalence, ankylosis secondary to AS is a rare condition. [4]. TMJ ankylosis is most commonly associated with trauma, local or systemic infection, and systemic rheumatic diseases including AS, rheumatoid arthritis, and psoriatic arthritis. Most cases show bilateral involvement of the TMJ. We report a case of unilateral bony ankylosis of the temporomandibular joint in a case of AS.

Case report

A 31-year-old male diagnosed with AS was referred for CT of the TMJ due to severe mouth opening limitation. The patient had a 16-year medical history of AS and sought assistance due to TMJ pain and incapacity to open his mouth. Clinical examination revealed decreased spine flexibility and impairment of upright postural alignment. Additionally, an interincisal distance of 2 mm on maximal mouth opening with inability to perform lateral and protrusive mandibular movements was registered.

Other recent imaging exams were available. Bony scintigraphy with ^{99m}Tc -MDP revealed tracer uptake along the spine, in the sacroiliac joints, in the right knee, and in the left TMJ. MRI with paramagnetic contrast medium revealed erosion of the left condyle and posterior slope of the articular eminence, and a mass of heterogeneous signal intensity between these structures. The left condyle also presented sclerosis/edema of the bone marrow and the disk could not be identified. No elective

concentration site of the paramagnetic contrast medium was observed (Fig. 1).

For the right side, sagittal and coronal CT images showed moderate erosion of the mandibular fossa, focal erosions on the articular surface of the condyle, and mild bony formation on the anterior aspect of the right condyle (Fig. 2a). On the left side, the images displayed markedly eroded condyle and mandibular fossa, and a bony mass resulting in ankylosis of the osseous components of the joint (Fig. 2b). Diagnosis of AS involving both TMJs, with associated bony ankylosis on the left side, was established.

The choice of treatment was taken by medical staff. The ankylotic mass on the left side and the diseased condyle were surgically removed. The TMJ disk was substituted by a silicone disk. Postoperative physiotherapy was prescribed. The result of the surgical procedure is shown on a postoperative panoramic image (Fig. 3).

After the procedure, a mouth opening improvement of 45 mm was found. During the patient's 12-month recall, this improvement was found to be reduced to 35 mm, but in a 2-year follow-up, it has stabilized at 35 mm.

Discussion

AS is a seronegative spondyloarthropathy (i.e., seronegative rheumatoid factor) that usually affects young adult men and has an important association with genetic predisposition, with expression of HLA-B27 genotype seen in most patients. A wide range of clinical presentations may be expected and quality of life may be severely threatened due to pain and disability [5].

Initial complaint generally consists of persistent low back pain. The disease usually jeopardizes the axial skeleton, first involving the sacroiliac joints symmetrically and then progressively reaching the spine from the lumbar to the cervical region. Around 30 % of the patients may present asymmetrical peripheral joints involvement, typically in the major proximal joints of the hips, shoulders, and knees. Involvement of small

Fig. 1 Degenerative alterations of the left TMJ displayed on axial and sagittal MR images

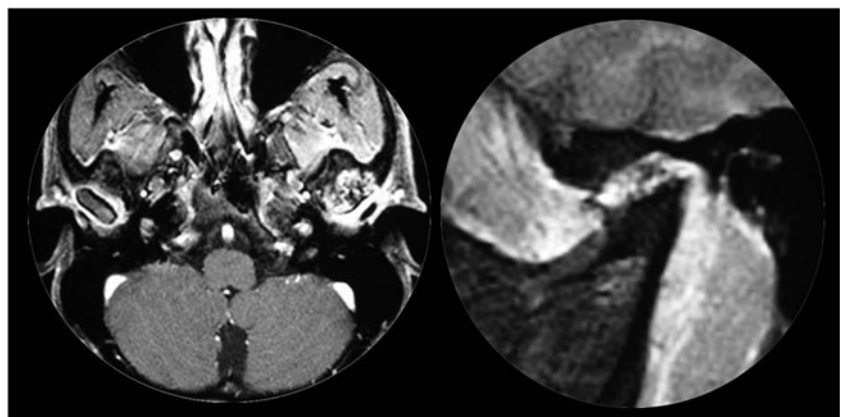
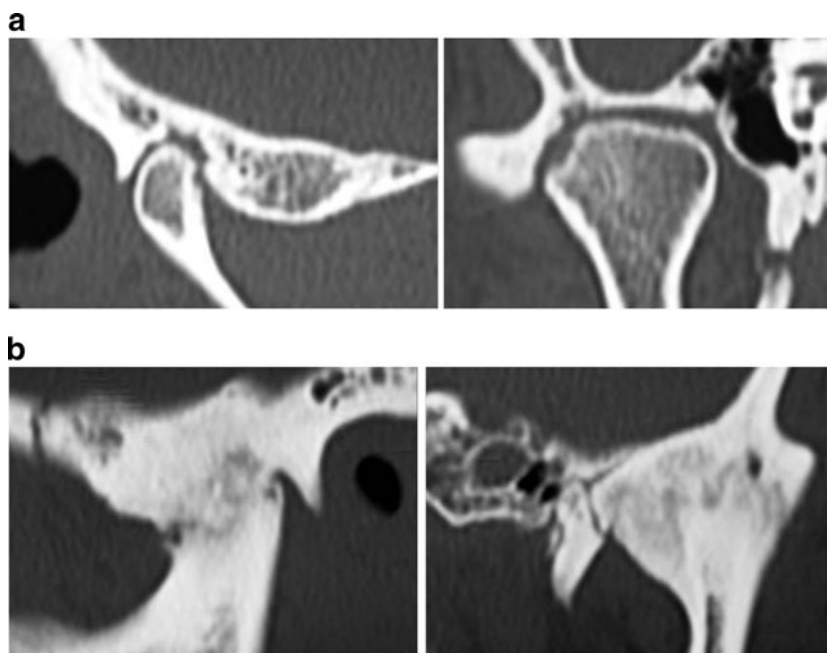


Fig. 2 **a** CT images, bone window, showing erosion of the articular osseous components and mild bony formation on the anterior aspect of the right condyle. **b** CT images, bone window, displaying a roughly eroded condyle and mandibular fossa and bony ankylosis of the left TMJ



peripheral joints is rarely observed. The cervical spine is the most susceptible site and may present a condition known as “bamboo spine” produced by ankylosis of vertebrae with restriction of neck movements [6].

Involvement of the TMJ may occur in patients with AS; however, the frequency reported in the literature is variable (15–54 %) and seems to be related to aspects of the populations studied and the methods used to assess TMJ features [7]. Resnick [8] reported that nearly one-third of long-standing AS patients presented TMJ alterations observed radiographically. The most common radiographic features are narrowing of the joint space, followed by condylar erosions, reduced mobility, osteophyte formation, demineralization, extensive sclerosis, and widening of the joint space [8]. Restricted mandibular movements are more common in AS patients than in healthy subjects [9].

Wenneberg et al. [9] found radiographic changes of the TMJ on panoramic radiographs in 25 % of AS patients, against 11 % in the control group. Condylar erosion was correlated with AS severity. In a later study, also through

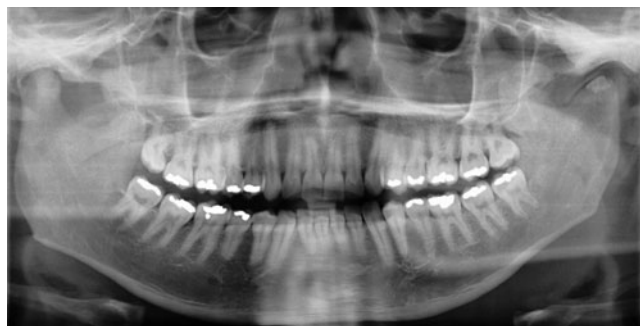


Fig. 3 Postoperative panoramic image showing the results of an aggressive resection of the ankylotic mass and silicone rubber disk replacement

panoramic radiographs, Wenneberg et al. [10] found radiographic changes more frequently in patients with rheumatoid arthritis (66 %), psoriatic arthritis (38 %), and AS (30 %) than in the control group (12 %).

Locher et al. [11] registered TMJ involvement in 22 % of AS patients through clinical and radiographic examination. TMJ involvement was considered when erosions and/or significant deformities of the condyle could be observed in combination with pain and/or limited mouth opening. The authors observed a tendency of patients with moderate or severe radiographic changes in the cervical spine to present TMJ changes.

Ramos-Remus et al. [7] observed that 49 % of AS patients reported subjective TMJ symptoms. Condylar erosions were statistically associated with longer disease duration, neck-related complaints, and anterior atlantoaxial subluxation. Erosions of the temporal bone were not common. Considerable variability in mandibular condyle mobility was observed among AS patients. On the other hand, erosions were not significantly associated with patient age, juvenile onset AS, activity index, TMJ symptoms, thoracic mobility, lumbar spine mobility, presence of HLA-B27, or presence of other peripheral joint involvements.

Major et al. [12] encountered a higher frequency and duration of headaches, TMJ pain, and painful jaw movements in AS patients compared to a control group. Disk displacement (62 %) and osseous degenerative changes (20 %) were more frequent in AS patients than in controls (34 and 6 %, respectively). Thus, the authors concluded that TMJ internal derangement and degenerative changes, as well as subjective pain complaints, are frequent among AS patients.

Ramos-Remus et al. [7] found that 90.9 % (10 out of 11) of the patients with severe AS presented alterations on MRI, including abnormal condylar shape (81.8 %), erosions

Table 1 Summary of clinical and imaging features of the current case and other previously reported cases of AS with TMJ ankylosis

Author (s)	Gender/age/history of AS (in years)	Side	Mouth opening	Pain	MRI/CT findings	Contralateral TMJ	Clinical features/other imaging modalities and joints
This study	M/31/15	Left	IO 2 mm	Present	Erosion of condyle and mandibular fossa Ankylotic bony mass	Focal erosions of condyle and moderate erosion of mandibular fossa	Impairment of upright position and reduced spine flexibility
[13]	M/40/n.i.	Bilat.	IO <10 mm No LM/PM	n.i.	Sclerosis/edema of the condylar bone marrow No visualization of articular disk (MRI) CT confirmed bilateral ankylosis	Mild bony formation on the anterior aspect of condyle	Scintigraphy revealed tracer uptake along the spine, sacroiliac joints, right knee, and left TMJ
[6]	M/20/6	Bilat.	IO 5 mm LM 1 mm	Present	–	–	Rigidity of cervical spine Malocclusion not present Classic aspect of “bamboo spine” on lateral radiograph of the cervical spine Bilateral sacroiliitis
[14] (in French)	M/48/18	Left	IO 10 mm No LM/PM	Present	Ankylosis seen on CT images	Condylar alteration, but no ankylosis seen on CT images	Malocclusion not present Ankylosis of C1–C2 observed on CT
[5]	M/28/10	Bilat.	IO 0 mm	n.i.	n.i.	–	Radiographs of dorsolumbar and lumbosacral suggestive of “bamboo spine” Extensive costovertebral ankylosis Severely restricted neck movements Unable to flex back or knees Involvement of spine and hips
[1]	M/59/30	Bilat.	IO <10 mm No LM/PM	Present	Complete bilateral obliteration of the TMJ space (CT) Irregular cortical margins and subcortical cystic lesions.	–	Fusion of C1–C2 Fused spine from the cervical to the lumbar region Right hip pain and stiffness Family history of ankylosing spondylitis
[14]	M/47/15	Bilat.	IO 5 mm	n.i.	Bone sclerosis and fusion seen on CT images	–	Morning stiffness in the dorsal and cervical spine Limited neck movements, with fixed flexion and lateral rotation of the cervical spine Kyphoscoliosis of the dorsolumbar spine with concavity toward the right side
[15]	F/24/12	Left	IO 2 mm.	Not present	–	–	–
[16]	M/34/14	Bilat.	IO 2–3 mm	n.i.	–	–	–

M male, F female, n.i. not informed, Bilat. bilateral, IO interincisal opening, LM lateral movement of the mandible, PM protrusive movements of the mandible

(45.5 %), and limited condyle translation (81.8 %). Thus, severe cases of AS usually affect the TMJ, and those patients may experience major TMJ problems (two cases of mild alterations were registered, while two and six cases of moderate and grossly abnormal shapes were found, respectively). However, even in the presence of gross imaging alterations, TMJ complaints may be absent in AS patients [7].

The basic principle in surgical release of ankylosis is the aggressive resection of the ankylotic mass. This technique is broadly termed gap arthroplasty and frequently employs the use of an interpositioning material to prevent recurrence [4]. In this case, a silicone rubber disk was used to substitute the disk, preventing ankylosis recurrence and restoring TMJ function. The surgical procedure was followed by aggressive postoperative physiotherapy to maintain optimal results as recommended for all surgical choices of procedure in ankylosis cases [4].

Ankylosis of the TMJ associated with AS is an extremely disabling affliction that impairs patients' ability to open their mouths, leading to further problems related to mastication, digestion, speech, and esthetics. In a recent review, Felstead and Revington [4] concluded that ankylosis of TMJ secondary to AS is a rare finding in the literature, especially with presentation of images from advanced diagnostic methods. We presented a case of AS with bilateral involvement of the TMJ and unilateral ankylosis of left condyle, showing different imaging modalities of the condition. Table 1 provides a summary of the clinical and imaging features of the current case, as well as other previously reported cases of AS with TMJ ankylosis.

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Conflicts of interest The authors state that there have no conflicts of interest.

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