CASE REPORT

Temporomandibular joint involvement in ankylosing spondylitis

Pallak Arora,¹ Janardhan Amarnath,² Setru Veerabhadrapa Ravindra,³ Mandeep Rallan⁴

¹Department of Oral Medicine and Radiology, Kalka Dental College Hospital and Research Centre, Meerut, Uttar Pradesh, India

²Department of Periodontics, Teerthankar Mahaveer Dental College & Research Centre, Moradabad, Uttar Pradesh, India

³Department of Oral Medicine & Radiology, Teerthankar Mahaveer Dental College & Research Centre, Moradabad, Uttar Pradesh, India

⁴Department of Pedodontics & Preventive Dentistry, Teerthankar Mahaveer Dental College & Research Centre, Moradabad, Uttar Pradesh, India

Correspondence to

Dr Mandeep Rallan,
drmandeepallan@gmail.com

SUMMARY

Frequency of temporomandibular joint (TMJ) involvement in patients with ankylosing spondylitis (AS) has varied from 4% to 35%. It is more common in men and produces generalised stiffness in involved joints. Clinician should be suspicious of AS when a patient reports with painful restricted movements of joint, neck or back and with no trauma history. Conventional radiographic methods have allowed the demonstration of TMJ abnormalities in patients with AS, but CT is necessary to establish joint space relations and bony morphology. We describe a case of severe AS with TMJ involvement in a 40-year-old female patient and demonstrated TMJ changes on CT. A CT was able to demonstrate articular cartilage changes, disc- and joint abnormalities. Thus, if conventional radiographs in a symptomatic patient with rheumatic diseases are unable to demonstrate changes, CT can provide valuable additional information of the changes in the TMJ.

BACKGROUND

Ankylosing spondylitis (AS) also known as Bechterew's disease or Marie-Strumpell disease is a chronic inflammatory disorder affecting predominantly the axial skeleton although peripheral joint involvement may be a significant feature.¹ In previous studies,² the male/female ratio is in favour of men ranging between 2.4:1 and 18:1. The disease affects synovial and cartilaginous articulations and the sites of tendon and ligament attachment to bone. The current diagnostic criteria include axial symptoms, limitation of spinal movement and radiological evidence of sacroiliitis.³ Immunological activity is suggested by the presence of human leucocyte antigen (HLA-B27) in over 90% of patients with AS.⁴

Arthritis of the TMJ with AS is less well recognised. Severe TMJ involvement was in fact present in Marie's original case⁵; however, since then, there have been only sporadic reports of its occurrence. The TMJ is involved in about 10–24% of cases. The commonest clinical symptoms are pain and tenderness in the TMJ region with limited jaw opening. The supposition that there is an arthritic involvement of the TMJ in patients with AS is by the radiological findings such as erosions or massive deformity of the condyle in combination with pain and/or restricted mouth opening.² Radiological examination of the TMJ is often unreliable in showing specific abnormalities, even when a number of different views are taken. This is improved by panoramic radiography in which both

joints may be compared directly and any asymmetry of opening could be seen.⁵ CT scanning represents the best imaging modality for the detection of osseous anatomy and pathology.² Typical findings are joint-space narrowing and condylar erosions.

Non-steroidal anti-inflammatory drugs (NSAIDs) are the first line of drugs and they effectively relieve symptoms. NSAIDs refractory patients are treated with second line drugs, for example, corticosteroid, disease-modifying anti-rheumatic drugs (DMARDs), pamidronate, etc.⁶ In patients with a painful, reduced mouth opening capacity, the treatment concept starts with the treatment of malfunctions using biofeedback and splint therapy as well as restorative and/or prosthetic rehabilitation.²

CASE PRESENTATION

A 40-year-old woman reported to the department with a history of limited mouth opening (figure 1) and pain in the right preauricular region since 1 year. She also gave the history of morning stiffness of jaws and complained of headache and watering of her eyes frequently. The patient had a medical history of AS diagnosed by an orthopaedician since 12 years. There was involvement of all the facet joints of spine resulting in thoracolumbar kyphosis so that she was unable to look up and unable to straighten herself while walking and the symptoms slowly worsened. She had no history of trauma or infection of the TMJ.

Physical examination revealed the limitation of spine mobility without neurological disturbances. There was marked neck flexion and forward thrusting of head (figure 2). The range of vertical mouth opening was 17 mm (figure 3). The unilateral

Figure 1 Restricted mouth opening.

To cite: Arora P, Amarnath J, Ravindra SV, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-009386

Figure 2 Neck flexion and forward thrusting of head.

preauricular tenderness was positive on palpation; with restricted condylar movement, there was no malocclusion.

INVESTIGATIONS

The relevant laboratory findings were raised erythrocyte sedimentation rate (ESR), that is, 56 mm, negative rheumatoid factor and positive HLA-B27.

Conventional radiograph of lumbar spine revealed sclerosis around apophyseal joint. Multiple bridging osteophytes (syndesmophytes) were seen in thoracolumbar vertebrae leading to the classic aspect of 'bamboo spine' appearance (figure 4). Pelvic radiography showed complete fusion of bilateral sacroiliac joints suggestive of grade 4 sacroiliitis. Bony erosions along with

Figure 3 Interincisal opening 17 mm.

Figure 4 Lumbar spine radiograph shows syndesmophytes bridging the vertebral bodies with a classic 'bamboo spine' appearance and sclerosis around the apophyseal joint.

sclerosis were seen in ischeal tuberosity and iliac crest suggestive of enthesopathy (figure 5).

Panoramic radiograph was not possible in our patient due to the approximation of chin to chest. A CT demonstrated TMJ changes, the condylar erosions on right side and narrowed joint space (figure 6).

TREATMENT

The patient was treated by a combination of conservative techniques including rest, reassurance and exercises for painful joint. She was prescribed sulfasalazine 1 g, hydroxychloroquine sulfate

Figure 5 Sacroiliitis. Conventional radiograph of the pelvis shows bilateral sacroiliitis; both the sacroiliac joints are fused completely. Extensive erosions along with sclerosis seen in ischeal tuberosity and iliac crest suggestive of enthesopathy.

Figure 6 CT view of temporomandibular joint (TMJ). (A) Right: erosive changes on the medial pole of the mandibular condyle and narrowed joint space. (B) Left: normal TMJ.

400 mg, indomethacin 75 mg, elemental calcium 500 mg and vitamin D₃ 250 IU for 1 week and was recalled after a week for follow-up.

OUTCOME AND FOLLOW-UP

Mouth opening increased from 17 to 30 mm. ESR was done after 15 days which had reduced to 30 mm and gradually unilateral preauricular pain disappeared.

DISCUSSION

The involvement of TMJ in AS appears to give rise to few serious symptoms until gross restriction of jaw movement has occurred. This contrasts with the acute pain and tenderness occurring in rheumatoid arthritis, which usually resolves spontaneously and only rarely gives rise to permanent restriction of jaw opening. Patients with TMJ involvement had, as might be expected, evidence of more extensive spinal disease and peripheral joint involvement than the remaining patients. The flexion deformity of the neck and fixed rigidity of the cervical spine in our patient caused some difficulty in assessing TMJ function due to the approximation of the chin to the chest, and it is interesting that marked neck flexion was present in three previous case reports.⁵ Davidson *et al*⁵ reported that the restricted mouth opening in the patients with AS could be due to the proximity of the chin to the neck. Few authors also reported that difficulty in opening the mouth may be correlated to the flattening and erosions of the mandibular condyle,² whereas Wenghoefer *et al*⁷ found that the limitation of jaw mobility in those patients might also be caused by an elongation of the mandibular coronoid process. In the present case, it is due to the approximation of chin to chest, which was due to the involvement of cervical spine as well. Approximately half of the affected patients had asymptomatic or unilateral involvement.¹ In the present case also, there was a unilateral involvement of TMJ.

Helenius *et al*⁸ reported that patients experienced pain, stiffness, headache and restricted movements in TMJ. Pain in the preauricular area is common. It has been suggested that

compression of retrodiscal tissue may be the cause of such pain. Wenneberg and Kopp (1982)⁹ reported that in the patient group, 13% has restricted mouth opening and 31% was tender to palpation of the TMJs compared with control group, with 4% having restricted mouth opening and 1% having preauricular tenderness. Similarly, our patient presented with restricted mouth opening, preauricular tenderness on right side, morning stiffness of jaws, headache, no clicking sounds and restricted condylar movements, in addition to all these findings, also complained of ocular symptoms which included frequent watering of eyes.

Ramos-Remus *et al*¹ found that AS had more variability in TMJ mobility than controls and showed increased frequency of condylar erosions, flattening, sclerosis and temporal flattening. It has been suggested in the past that in AS, radiological changes in TMJ occur at a relatively late stage.¹ Resnick (1974)¹⁰ reported that 32% of his sample of 25 consecutive patients with long-standing AS had tomographic TMJ abnormalities. The most common feature was joint space narrowing, followed by erosions, reduced mobility, osteophyte formation, excessive sclerosis and extensive erosion.¹ In the present case, a CT revealed condylar erosion and abnormal shape of condyle on the affected side. Panoramic radiography was not possible in our patient due to the approximation of chin to chest; it is also reported by Locher *et al*² that in 3 out of 50 patients an orthopantomograms (OPG) was not possible because of the stiffness of the patients. Conventional radiography is only an approximate diagnostic method for the examination of TMJ. A better radiographic method for the condyle, the joint space and the articular fossa is CT, especially if the prevalence of early radiographic signs is to be investigated.² Condylar erosions were reported to be correlated with the severity and duration of AS and are diagnostic of degenerative change in osseous tissue.¹

It is difficult to judge a radiographic change as being of arthrotic or arthritic origin, even if it is well-known that arthritis can lead to arthrosis by the traumatic influence of movement on a cartilage impaired by inflammation, resulting in a postarthritic arthrosis.² Therefore, it is not possible to state whether TMJ involvement in AS is due to arthritic or arthrotic changes. The first radiographic sign in arthritis is erosion, which arises from the osteolysis of the subchondral bone. Exophytes, cysts, flattening of the condyle and/or the fossa are signs of arthrotic changes. They occur quite often in the general population.² The mechanism by which TMJ involvement is seen in patients with AS is unclear; however, there are two potential mechanisms for the pathogenesis of TMJ involvement in AS. One could involve the destruction of the capsular or disc attachment, resulting in internal derangement and subsequent degenerative joint diseases. Alternatively, there could be a primary synovitis with direct breakdown of the articular surfaces. Internal derangement would then result from articular surface changes and not precede them. Hypermobility was the most common finding. Destruction of capsular attachment would result in hypermobility. Hypomobility could be due to disc derangement or fibrosis of the capsule.

Cervical dysfunction, with neck complaints as a presenting symptom, and atlantoaxial subluxation were significantly associated with advanced TMJ involvement as evidenced by erosions. AS causes debilitating postural changes with forward thrusting of the head.¹ Postural imbalance of neck may affect the function of the masticatory system and TMJ involvement may be a result of this abnormal posture, rather than of the disease itself, but this hypothesis appears unlikely. If the involvement was through the dysfunction of masticatory muscles, most

patients would also have reported facial pain; there was no facial pain in the present case as well.

Treatment is multidisciplinary and may involve many specialists from the outset. Physiotherapy, preferably with a specific AS exercise programme, is essential in helping to maintain posture and prevent fusion of the spine. Anti-inflammatory medications are still a major part of symptomatic treatment. DMARDs such as sulfasalazine, methotrexate, hydroxychloroquine sulfate, etc can be effective in peripheral joint inflammation. Anti-TNF- α therapies are the most exciting therapeutic area to date. Infliximab and etanercept are now licensed for the treatment of AS.⁶

Learning points

- ▶ A CT serves as an important diagnostic modality for temporomandibular joint (TMJ) disorders.
- ▶ Conservative techniques including rest, reassurance, physiotherapy and non-steroidal anti-inflammatory (NSAID) medication serve as the first line of treatment for a majority of TMJ disorders.
- ▶ By improving joint mobility and her ability to eat, oral physicians can improve quality of life of patients in a debilitating disease like ankylosing spondylitis, the patient got a new hope and a will to fight and win over the disability caused by the disorder, which had previously appeared impossible for the patient.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Ramos-Remus C, Major P, Gomez-Vargas A, *et al*. Temporomandibular joint osseous morphology in a consecutive sample of ankylosing spondylitis patients. *Ann Rheum Dis* 1997;56:103–7.
- 2 Locher MC, Felder M, Sailer HF. Involvement of the temporomandibular joints in ankylosing spondylitis (Bechterew's disease). *J Craniofac Surg* 1996;24:205–13.
- 3 Deesomchok Q, Tumrasvin T. Clinical study of Thai patients with ankylosing spondylitis. *J Clin Rheumatol* 1985;4:76–82.
- 4 Manemi RV, Fasanmade A, Revington PJ. Bilateral ankylosis of the jaw treated with total alloplastic replacement using the TMJ concepts system in a patient with ankylosing spondylitis. *Br J Oral and Maxillofac Surg* 2009;47:159–61.
- 5 Davidson C, Wojtulewski JA, Bacon PA, *et al*. Temporo-mandibular joint disease in ankylosing spondylitis. *Ann Rheum Dis* 1975;34:87–91.
- 6 Ghosh A, Kole A, Devi GI, *et al*. Treatment of ankylosing spondylitis with special reference to biologics: single centre experience. *J Indian Rheumatol Assoc* 2004;12:54–7.
- 7 Wenghoefer M, Martini M, Allam JP, *et al*. Hyperplasia of the coronoid process in patients with ankylosing spondylitis (Bechterew disease). *J Craniofac Surg*. 2008;4:1114–18.
- 8 Helenius LMJ, Hallikainen D, Helenius I, *et al*. Clinical and radiographic findings of the temporomandibular joint in patients with various rheumatic diseases. A case control study. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2005;99:455–63.
- 9 Wenneberg B, Kopp S. Subjective symptoms from the stomatognathic system in ankylosing spondylitis. *Acta Odontol Scand* 1982;40:215.
- 10 Resnick D. Temporomandibular joint involvement in ankylosing spondylitis. Comparison with rheumatoid arthritis and psoriasis. *Radiology* 1974;112:587–91.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow