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Temporomandibular joint ankylosis suspected to be associated with ankylosing spondylitis based on cervical computed tomography images: A pictorial essay

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ABSTRACT

This report showed a case of temporomandibular joint (TMJ) ankylosis suspected to be associated with ankylosing spondylitis based on the observation of bony ankylosis of the cervical spine on computed tomography (CT) images. A 53-year-old man presented with a chief complaint of difficulty in opening his mouth. His medical history indicated that in his 20 s, he became aware of the difficulty in moving his neck. CT revealed marked osteoarthritic changes in the right mandibular condyle, suggesting fibrotic TMJ ankylosis. In addition, bony ankylosis of the cervical vertebral body and facet joints from the axis (C2) to C5 in continuity was observed. CT of the entire spine also showed bony deformity of the sacroiliac joints and bony ankylosis. Based on these findings, ankylosing spondylitis was suspected. The possibility of an ankylosing spondylitis complication should be considered in cases of TMJ ankylosis if bony ankylosis of the cervical spine is observed. ***(Imaging Sci Dent 2024; 54: 201-6)***

KEY WORDS: Spondylitis, Ankylosing; Temporomandibular Joint; Computed Tomography, X-Ray

Ankylosing spondylitis is a rheumatoid factor-negative spondyloarthritis that causes chronic systemic inflammatory disorders and is generally characterized by the inevitable occurrence of sacroiliitis.[1] It is more common in male sex than in female sex, and more than 80% of cases occur before 30 years of age.[1,2] Ankylosing spondylitis causes stiffness, decreased mobility, and pain in the lower back; however, nonspecific findings often make diagnosis difficult.[1,3] Initial complaint typically involves persistent low back pain.[1,3] The disease usually jeopardizes the axial skeleton, first involving the sacroiliac joints symmetrically and subsequently progressing to the spine from the lumbar to the cervical region.[1,3,4] Immunological activity is indicated by the presence of human leucocyte antigen (HLA-B27) in more than 90% of patients with ankylosing spondylitis.[5,6] Ankylosing spondylitis is a disease of high diagnostic value that is associated with multiple complications such as uveitis, psoriasis, and inflammatory bowel disease.[6]

Peripheral joint involvement can also occur in patients with ankylosing spondylitis. The prevalence of temporomandibular joint (TMJ) involvement in ankylosing spondylitis ranges from 4% to 59%.[7-10] TMJ symptoms include pain, stiffness, and limited jaw movement.[11,12] Despite these high prevalence rates, TMJ ankylosis secondary to ankylosing spondylitis is rare,[13] and some cases of TMJ ankylosis in patients with ankylosing spondylitis have been previously reported.[4,11,13-17] Radiographic findings of TMJ ankylosis include narrowing of the joint space, erosion of the condylar facets, and osteophyte formation.[10-12] Although the pathogenesis of the TMJ involvement remains unknown, altered joint mobility and association with atlantoaxial subluxation suggest primary involvement of the capsular and disk attachment.[18] It is difficult to distinguish between normal TMJ ankylosis and secondary TMJ anky-

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losis associated with ankylosing spondylitis, which cannot be definitely proven. Treatment is based on extensive surgical resection, and intensive and regular jaw opening training is essential for a prolonged postoperative period.[14] In all the previous cases,[4,11,13-17] ankylosing spondylitis was first diagnosed, followed by the later diagnosis of TMJ ankylosis attributed to the development of TMJ-related symptoms. In this report, we describe the case of a patient with no history of ankylosing spondylitis, who presented with a chief complaint of difficulty in mouth opening. A computed tomography (CT) scan of the TMJ revealed bony ankylosis of the cervical spine, which was suspected to be ankylosing spondylitis.

Case

This case report was approved by the Ethics Committee of our institutional review board (No. 33062). A 53-year-old man presented with a chief complaint of difficulty opening his mouth. His medical history indicated that in his 40s, he became aware of experiencing difficulty in opening his mouth. He was diagnosed with TMJ disorder at a primary dental office, where he started receiving treatment; however, he later self-discontinued it. Recently, during treatment for caries, limited mouth opening was observed for which he was referred to Tohoku University Hospital. His medical history included hypertension (since his late 40s) and depression (since he was 21 years old). In his 20s, he became aware of difficulty moving his neck. He had self-determined that the side effects of the medication were causing stiffness in his neck and cervical area, and thus he discontinued taking the medication and visiting the psychosomatic medicine clinic by himself for a certain period of time. Imaging examination of his neck was not performed at the time. A physical examination revealed a maximum mouth opening of 20 mm (both of self-opening range and forced opening range) and limited mandibular condylar mobility. Laboratory data revealed a mildly elevated C-reactive protein level but no other abnormal values of other immunological markers. The presence of human leukocyte antigen (HLA) B27 was not tested.

Panoramic radiography (Veraviewepocs; Morita Corp.,

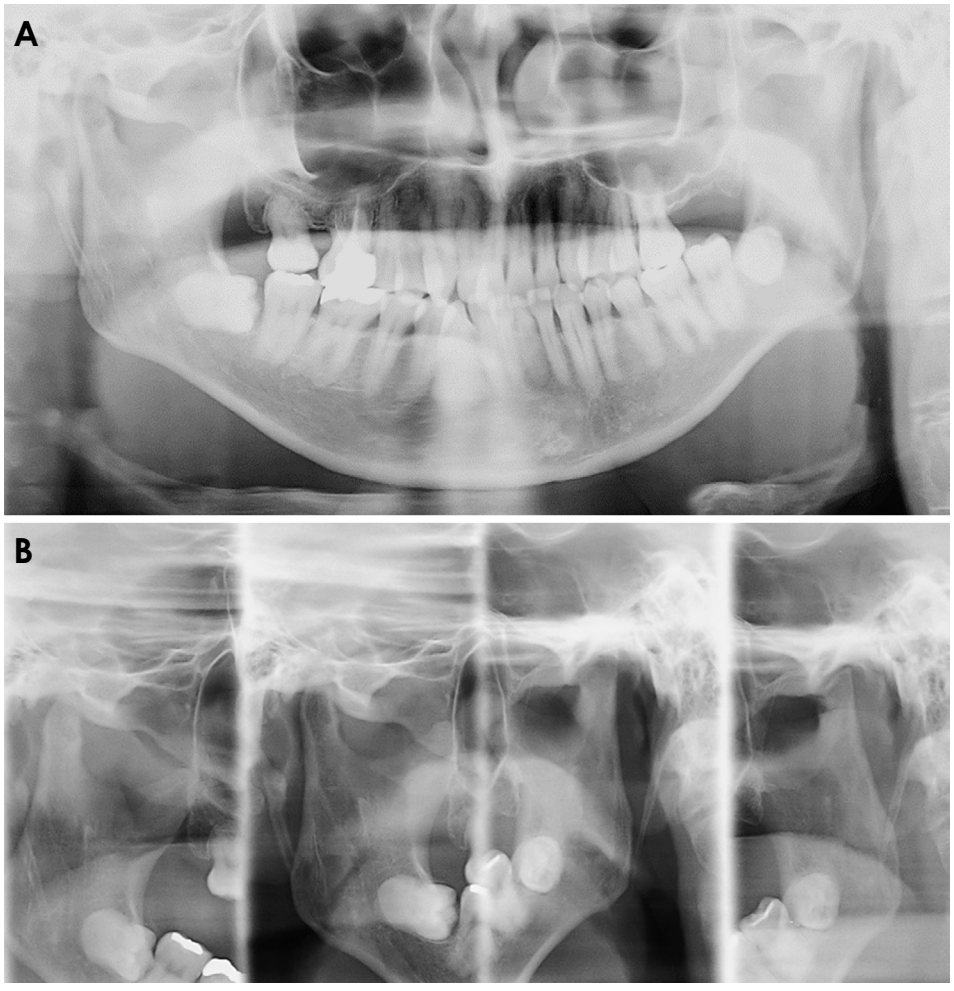

Fig. 1. A. Bilateral mandibular condyle shows osteoarthritic change on the panoramic radiograph. B. Temporomandibular joint panoramic mode radiograph shows the restricted bilateral mandibular condyle movement.

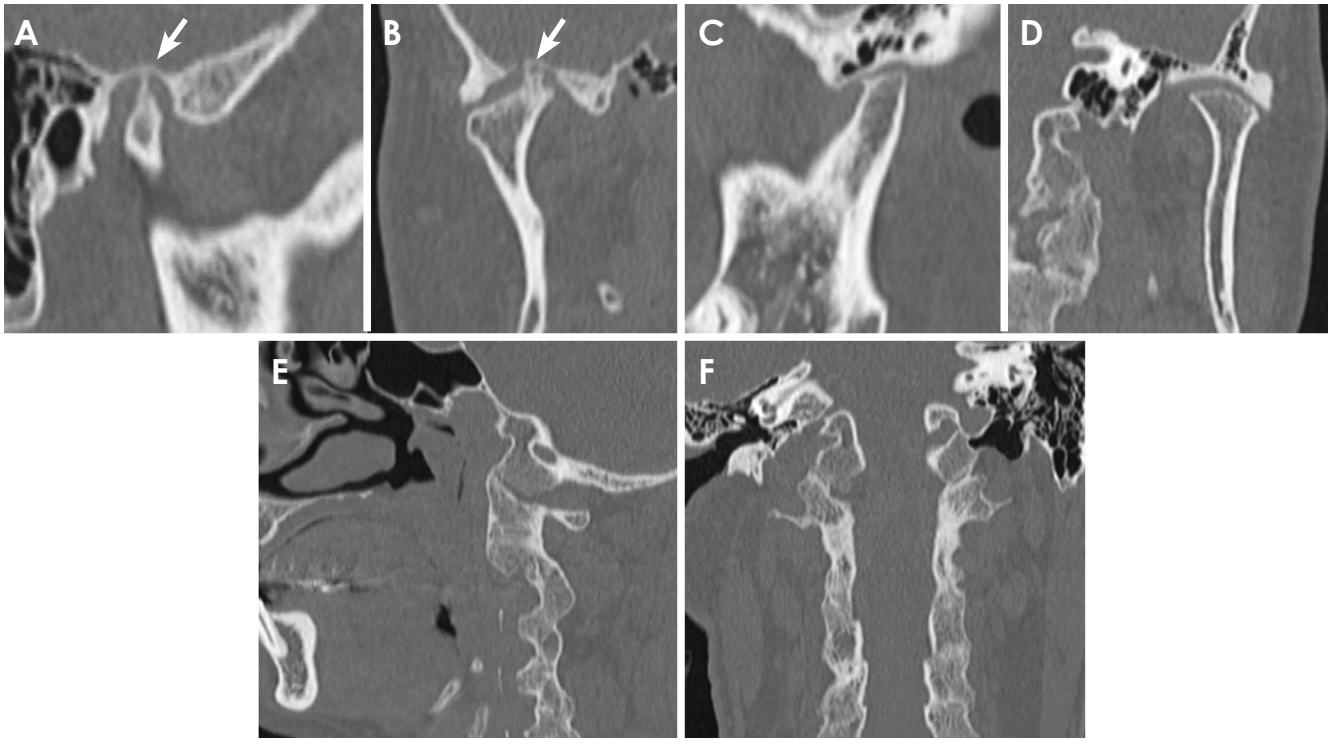

Fig. 2. Sagittal (A) and coronal (B) computed tomographic (CT) images of right temporomandibular joint (TMJ) show marked osteophyte formation. The right TMJ space is markedly narrowed, and the top of condyle is in contact with the mandibular fossa (arrows). C. Sagittal (C) and coronal (D) left TMJ CT images show flattening of the condylar head. Bilateral TMJs show bone sclerotic change. Sagittal (E) and coronal (F) cervical vertebral body CT images show the facet joints of the cervical vertebrae are osseous continuous in C2-5.

Kyoto, Japan) showed erosion of the right mandibular condylar surface, narrowing of the bone morphology toward the condylar head, and flattening of the left condyle. Bilateral mandibular condylar movements were restricted (Fig. 1). CT (SOMATOM Definition Flash; Siemens Medical Solutions, Erlangen, Germany) showed marked osteophyte formation in the right condylar head and flattening of the left condylar head. The right TMJ space was markedly narrowed, and the top of the condyle was in contact with the mandibular fossa, suggesting fibrotic TMJ ankylosis. In addition, bony ankylosis of the cervical vertebral body and the facet joints from the axis (C2) to the lower end of the imaging range (C5) in continuity was observed (Fig. 2). Magnetic resonance imaging (MRI) (Signa HDxt; GE Healthcare, Fairfield, CT, USA) showed marked osteophyte formation in the right condylar head, similar to the CT findings, and medial disk displacement was suspected in the right TMJ. In addition, bony ankylosis of the cervical vertebral body and facet joints from C2 to C7 was observed (Fig. 3). Ankylosing spondylitis as a complication of TMJ ankylosis was suspected, and the patient was referred to an orthopedic surgeon.

Orthopedic examination revealed that the cervical spine had no range of motion in forward flexion, backward flexion, lateral flexion, and rotation. CT of the entire spine also showed bony deformity of the sacroiliac joints and some bony ankylosis. Bony ankylosis of the facet joints of the thoracic vertebrae was also observed (Fig. 4). An orthopedic diagnosis of ankylosing spondylitis was suspected based on clinical and CT findings.

The patient was carefully followed up in the orthopedic department to monitor the progression of his symptoms and the bony deformities. He was diagnosed with fibrotic TMJ ankylosis based on the markedly reduced self-opening range of 20 mm/forced opening range of 20 mm, no increase in the amount of forced opening, restricted range of movement of the mandibular condyle on physical examination and imaging findings, and MRI findings of contour obscuration rather than displacement of the disk. Bilateral TMJs contractures could not be ruled out. Based on the above findings, the patient was recommended to undergo TMJ arthroscopic surgery to remove fibrotic adhesions and to improve the opening difficulties. Informed consent for the operation was obtained from the patient.

TMJ arthroscopic surgery was performed. Diagnostic arthroscopy was performed by puncturing the superior

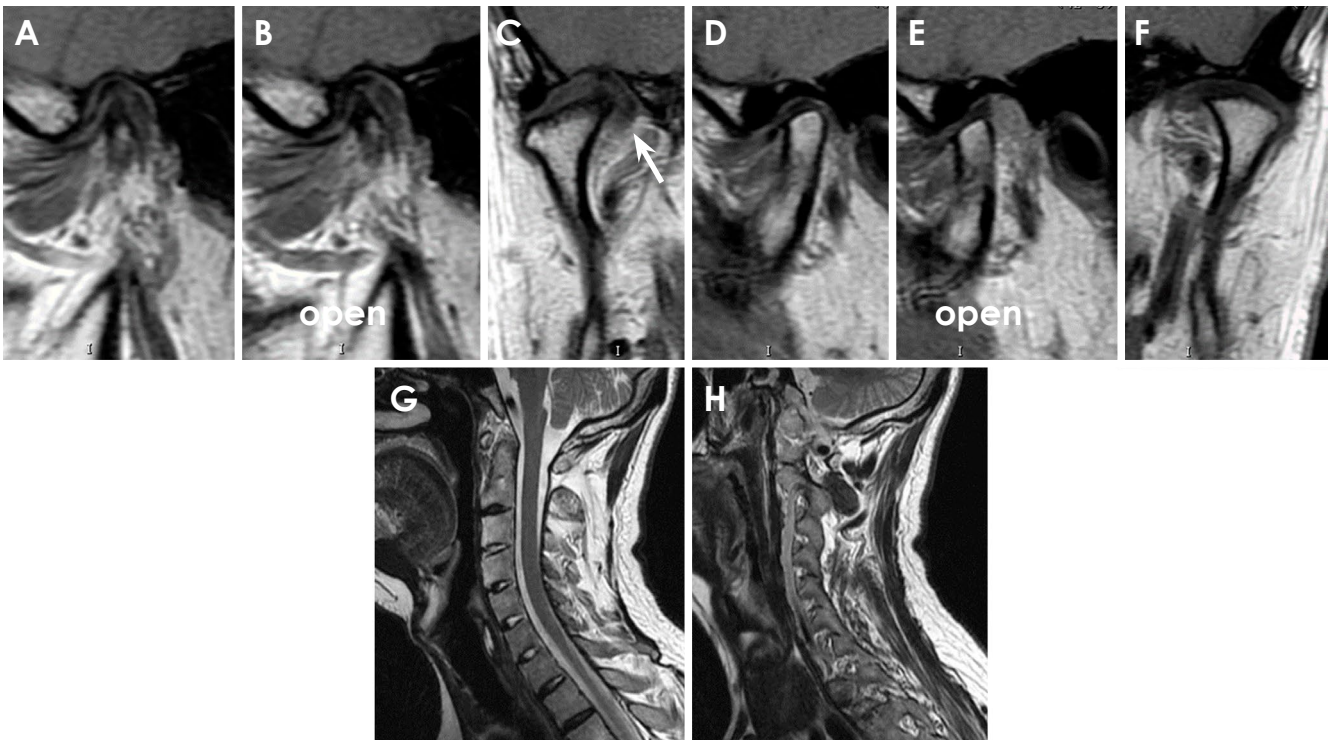

Fig. 3. Close sagittal (A), open sagittal (B), and close coronal (C) right temporomandibular joint (TMJ) MR images show marked osteophyte formation and medial disk displacement on proton density weighted image. Close sagittal, open sagittal (E), and close coronal (F) left TMJ images show flattening of condylar head and no disk displacement on proton density weighted image (spin echo technique; repetition time, 1600 ms; echo time, 20 ms). Sagittal vertebral body image (G) and sagittal cervical facet joints of the cervical vertebrae image (H) show osseous continuous in C2-7 on T2 weighted image (fast spin echo technique; repetition time, 3000 ms; echo time, 106 ms).

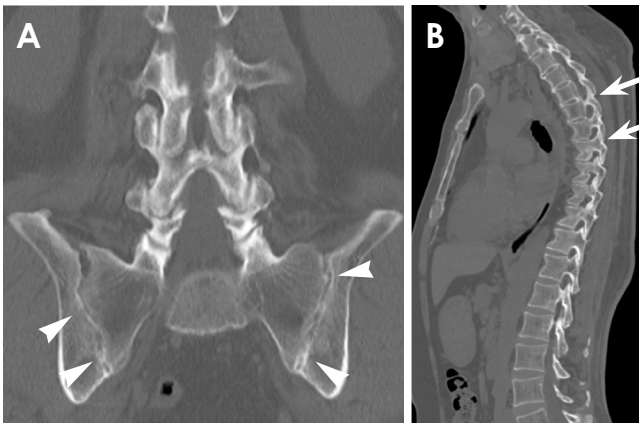

Fig. 4. A. Coronal bone computed tomography (CT) image. B. Sagittal bone CT image. The vertebrae are squared throughout the spine. Ossified osteophytes are evident in the cervicothoracic spine. The bilateral sacroiliac joints are partially ankylosed (arrowheads). Bony ankylosis of the facet joints of the thoracic vertebrae is present (arrows).

joint space and inserting an arthroscope. Fibrotic adhesions were observed intraoperatively, and a trocar was inserted through the puncture site to dissect the fibrotic adhesions and ensure mobility of the mandibular condyle, following which closed sutures were placed. Histopathological analysis showed fibrous tissue growth. Based on the intraoperative fibrotic adhesions and histopathological fibrotic tissue hyperplasia, the diagnosis of fibrotic temporomandibular joint ankylosis was confirmed.

The patient showed improvement in mouth opening at 3 months postoperatively with an opening of 34 mm, and imaging findings showed gliding movement of the mandibular condyle (Fig. 5). In the subsequent 2-year follow-up, a maximum mouth opening remained stable over 30 mm.

Discussion

Ankylosing spondylitis is generally associated with sacroiliitis, and the spondylitis often progresses from the lumbar spine to the cervical spine.[1,3,4] The etiology of ankylosing spondylitis is unknown; however, HLA-B27 is genetic factor known to be strongly associated with the disease. HLA-B27 is detected in approximately 90% of patients with ankylosing spondylitis.[5,6] In Japan, the fre-

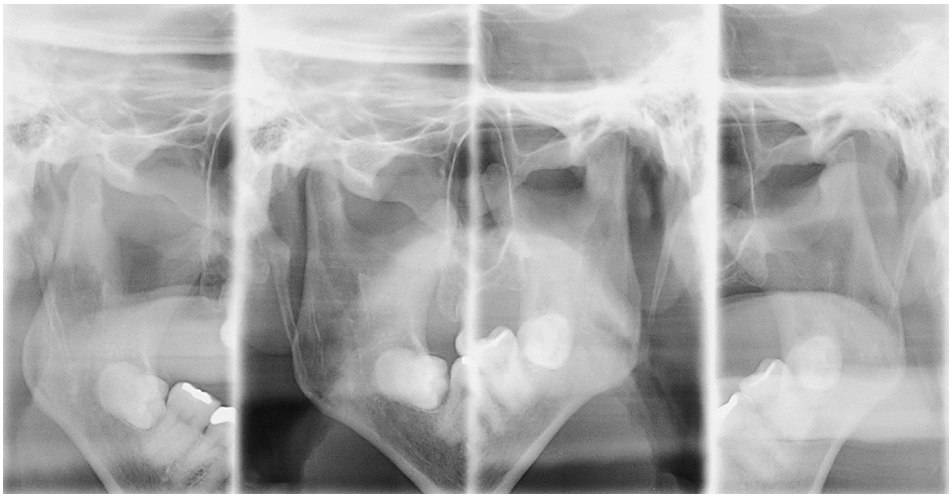

Fig. 5. Three-month postoperative panoramic temporomandibular joint view shows recovery of the sliding movement of the bilateral mandibular condyle.

quency of HLA-B27 in the general population and the prevalence of ankylosing spondylitis are much lower than those reported in Western countries.[19,20] The patient in the present case had advanced bony ankylosis of the cervical spine, which is somewhat atypical for ankylosing spondylitis; however, the presence of HLA-B27 was not tested in this case. In addition to cervical spine ankylosis, the imaging findings in this case showed sacroiliitis, leading to the diagnosis of suspected ankylosing spondylitis based on the criteria.[1,3]

In previous reports of TMJ ankylosis in patients with ankylosing spondylitis,[4,11,13-17], all the patients had a history of ankylosing spondylitis when they were diagnosed with TMJ ankylosis. However, in the present case, ankylosing spondylitis was not identified at the time of the initial medical history interview. Although the patient became aware of stiff neck symptoms in his 20 s, he was not diagnosed with ankylosing spondylitis at the time. The patient reported his past "cervical symptoms in his 20 s" after a CT scan of the cervical spine revealed bony stiffness of the cervical vertebrae. As follow-up of ankylosing spondylitis cases is important for evaluating the severity of disability and the development of complications due to the progression of bony ankylosis,[1,6] the ability to diagnose ankylosing spondylitis in this case without being limited to a diagnosis of TMJ alone is very significant. This case indicates that ankylosing spondylitis should be considered if a patient's TMJ-related symptoms include difficulty with mouth opening suspected to be due to bony ankylosis or if bony ankylosis of the cervical spine is detected. In addition, it is necessary for clinicians to be conversant with the symptoms of spondyloarthritis and the presence of other related complications.

In a review of the imaging findings of TMJ ankylosis in ankylosing spondylitis, Wenneberg et al.[10] reported that approximately one-third of patients with long-term ankylosing spondylitis show bony deformities of the TMJ on radiographic images. The most common radiographic features are narrowing of the joint space, condylar erosion, reduced mobility, osteophyte formation, and extensive sclerosis. In addition, restricted mandibular movement is more common in patients with ankylosing spondylitis than in healthy individuals.[10,12,14] In a previous study, histopathological examination of TMJ disks and posterior attachments removed from 15 patients with chronic arthritic diseases such as rheumatoid arthritis and psoriatic arthropathy, including five patients with ankylosing spondylitis, revealed more pronounced changes in vascular proliferation, perivascular cellular infiltration, inflammatory cells, and soft tissue fibrosis in patients with chronic arthritic disease than in those with normal osteoarthritis of the TMJ; destruction of the disk was also a prominent finding in patients with chronic arthritic disease.[21] This suggested that patients with chronic arthritic disease might develop osteoarthritis of the TMJ through a mechanism somewhat different from that of normal osteoarthritis of the TMJ. On the other hand, it has been reported that postural changes occur due to cervical spine stiffness and as secondary effects of muscle disorders.[7] It is well known that arthritis can lead to arthrosis through the traumatic influence of movement on a cartilage impaired by inflammation, resulting in post-arthritic arthrosis. However, it is difficult to diagnose a radiographic change as being of arthrotic or arthritic origin. Therefore, it is not possible to determine whether TMJ involvement in ankylosing spondylitis is due to arthritic or arthrotic changes.

In conclusion, this report described an interesting case of TMJ ankylosis suspected to be associated with anky-

losing spondylitis based on cervical CT images. The possibility of an ankylosing spondylitis complication should be considered in cases of TMJ ankylosis if the patient's symptoms include difficulty with mouth opening due to suspected bony ankylosis and bony ankylosis of the cervical spine is observed.

Conflicts of Interest: None

Acknowledgments

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