CASE REPORT

Synovial chondromatosis in the temporomandibular joint: a case with typical imaging features and pathological findings

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ABSTRACT. Synovial chondromatosis of the temporomandibular joint is a rare benign joint disorder that has been reported in only a few studies. However, we recently encountered a pathologically proven case of this disorder. This case also showed the typical imaging findings on panoramic radiographs and on CT and MR images. Therefore, we report this case and the imaging and pathological findings.

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Synovial chondromatosis (SC) is an uncommon articular disorder characterised by synovial metaplasia with intra-articular proliferation of cartilaginous nodules originating from the synovial membrane [1–3]. This disorder usually affects large joints and is rarely observed in the temporomandibular joint (TMJ). SC was originally described by Ambroise Pare in 1558 and the involvement of the TMJ was described in 1933 by Auhausen [3, 4]. CT, MRI and arthroscopy facilitate the clinical diagnosis of SC; however, pathological analysis is mandatory to confirm the disease [2]. To our knowledge, most cases were assessed on the basis of CT or MR images and few studies have reported findings from all imaging modalities, including plain radiography. We report a case of SC in the TMJ, which was confirmed by histopathological analysis, and present the typical imaging findings, including CT, MR and plain radiography findings.

Case report

A 49-year-old man who had been experiencing pain on the right TMJ region for several years was referred to Wonkwang University Hospital, Iksan, Jeonbuk, Korea, because of acute exacerbation of symptoms 5 days previously. He complained of pain and a clicking sound while opening his mouth.

Physical examination revealed tenderness and swelling in the TMJ region; he could not open his mouth more than 27 mm because of the pain.

A panoramic radiograph showed multiple calcified loose bodies around the right mandibular condyle (Figure 1). Axial CT images showed widening of the TMJ space and multiple calcifications around it (Figure 2a). The images

also showed bony erosion with sclerotic changes in the adjacent articular eminence and glenoid fossa (Figure 2b).

MRI was subsequently performed and showed a soft tissue mass expanding into the joint space and multiple nodules in the intra-articular soft tissue mass. The intra-articular mass had high signal intensity (SI) and the small nodules had low SI on T_1 weighted images (WI). Anterior displacement of TMJ disc was also observed. Gadolinium-enhanced T_1 WI showed wall enhancement representing synovial thickening (Figure 3a,b).

A presumptive radiological diagnosis of synovial chondromatosis was made.

Surgical exploration of the mass via a preauricular approach was performed under general anaesthesia.

Figure 1. Panoramic radiograph shows calcified nodular lesions (arrows) in the right temporomandibular joint space.

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Figure 2. (a) Axial CT image shows multifocal calcified loose bodies (arrow) within a low-attenuated soft tissue mass anterior to the right mandibular condyle. (b) Coronal CT image also shows loose bodies (arrow). It shows bony erosion of glenoid fossa (arrowhead) and widening of joint space but does not show extra-articular extension. (c) Follow-up CT obtained after 10 months confirms complete removal of intra-articular mass and calcifications and the absence of recurrence.

Multiple loose bodies were observed within the joint space (Figure 4a) and histopathological assessment showed chondroid metaplasia with thick fibrosis and calcification (Figure 4b). Therefore, we made a confirmative diagnosis of synovial chondromatosis.

The mass was completely removed and the patient's symptoms improved. After 10 months, the patient underwent follow-up CT and no residual or recurrent lesion was observed (Figure 2c).

Discussion

SC is an uncommon, benign, monoarticular arthropathy that is characterised by the formation of cartilaginous bodies in the synovial membrane. This disease usually affects large joints [1–3, 5]. SC in the TMJ is rare and may show nuclear atypia, which indicates malignancy [6]. It occurs more often in women, which is opposite to the trend for SC in other joints, between the

fourth and sixth decades of life. The clinical features of SC in the TMJ are pain, swelling, limitation of the movements of the jaw and crepitation on mouth opening. The presence of cranial nerve dysfunction indicates that the disease has reached the advanced stage [5, 7–9].

The pathogenesis of SC is unknown. Some contributing factors, such as previous trauma, parafunctions, degeneration, inflammatory disease and infections, have been suggested but these do not seem to be the main causes of SC [2, 7, 10, 11]. Some previous studies have suggested that fibroblast growth factor 2 and fibroblast growth factor receptor 1 play important roles in the pathogenesis of SC [12].

On the basis of histopathological findings, SC can be divided into the following three stages:

1. Initial stage: in this stage, the lesions include metaplasia of the synovial membrane with proliferation of undifferentiated cells. Detached loose bodies are not observed at this stage.

Figure 3. (a) T_1 weighted (W) sagittal image shows multiple loose bodies, most of which have small and low signal intensity (SI) within a soft tissue mass (arrows). The soft tissue mass shows high SI because of fluid collection in it which represents expanded articular cavity. (b) Post-contrast T_1 W coronal MR image shows a soft tissue mass (arrows) expanding into the right temporomandibular joint space. This mass shows peripheral wall enhancement representing thickened synovium. Further, masticator space and pterygoid muscle with solid components are enhanced because of the metaplasia of the synovial tissue.

Figure 4. (a) Numerous small calcified nodules with variable sizes are retrieved from the joint space. (b) Histopathological ($\times 40$) analysis shows small, metaplastic cartilaginous nodules of varying size with calcifications.

2. Transitional stage: this stage shows the presence of progressive metaplasia in the synovial membrane along with formation of loose bodies that contain active chondrocytes partially covered by the synovial membrane.
3. Advanced stage: no intrasynovial metaplastic activity is detected in this stage; only detached loose bodies are observed [13].

SC can also be divided into two forms according to the cause of the disorder: primary and secondary. In the primary form, metaplastic changes develop in the synovium and cause cartilaginous foci to develop within the synovial membrane. These foci enlarge, detach and ossify in the synovial cavity. The secondary form occurs following trauma or other arthropathies. Fragments of cartilaginous or bony tissue dislodge and are nourished by the synovial fluid. The primary form is more aggressive and tends to recur. In contrast, the secondary form is usually slow and has a chronic presentation. Moreover, in the secondary form, the loose bodies are more uniform and show less cellular atypia than that observed in the primary form [14].

SC is a benign disease, but it does not respond to non-surgical treatment and does not show spontaneous resolution. Moreover, it has the potential to invade the intracranial structure when involved with the TMJ [5, 8]. Therefore, early and accurate diagnosis is very important for selecting an appropriate treatment and for a better prognosis [15]. Imaging tools for diagnosis include conventional radiography, CT, MRI and arthroscopy. Although the definitive diagnosis is usually made on the basis of arthroscopic or open examinations, in recent reports, MRI has also been shown to provide an imaging perspective of the pathological nature and facilitate a pre-operative verdict [4]. The main imaging features of SC in the TMJ are as follows: widening of the joint space, soft tissue swelling, irregular surfaces of joints and multiple calcified loose bodies. Coronal CT images are particularly good for detecting loose bodies in the joint spaces [1–4]. Although many conditions present with loose bodies within the TMJ, one notable difference is that patients with SC have more than 10 loose bodies and these are packed in the soft tissue mass. This appearance resembles that of grains of rice in sushi [8, 16]. MRI

is useful for revealing boundaries, assessing internal derangement and detecting intracranial extension. The detection of skull-base changes is important because it indicates a more aggressive course that can potentially destroy the skull base and intrude into intracranial structures. Therefore, pre-operative CT and MRI are very useful for early detection of these changes and are beneficial in improving the prognosis of the patients [1].

SC does not spontaneously resolve and may show recurrence. Therefore, it requires appropriate treatment and post-treatment follow up. The treatment options for SC are surgery and arthroscopy. Arthroscopy is a less invasive technique, but it is difficult to retrieve the intra-articular loose bodies using arthroscopy. In some cases, the lesion may show extracapsular and intracranial extension and SC of the TMJ has the potential to undergo malignant transformation. Therefore, the treatment of choice is surgical exploration [2, 5, 7]. Surgical treatment should be followed by regular follow-up examinations and MRI can be useful in the follow-up period. These examinations should be conducted at intervals of 6 months initially and once a year thereafter [7].

SC of the TMJ has been reported in some studies. However, to our knowledge there is no report that presents the pathological findings along with the typical imaging findings from all imaging modalities. If physicians were aware of all these findings, the diagnosis of this condition would be greatly facilitated, thereby ensuring prompt and adequate therapy and improving the prognosis of the patients.

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