

## 2

# Synovial Chondromatosis in the Temporomandibular Joint: Report of Two Cases

단국대학교 치과대학 구강악안면외과학교실

하 용 찬, 김 철 환

## ABSTRACT

### Synovial Chondromatosis in the Temporomandibular Joint: Report of Two Cases

Department of Oral and Maxillofacial Surgery, College of Dentistry, Dankook University  
Yong-Chan Ha, DDS, Chul-Hwan Kim, DDS, MS, PhD

#### Background

Synovial chondromatosis(SC), a proliferative disorder of the synovial membrane. The etiology or cause of SC remains unclear. SC usually occurs in large articular joints such as knee, hip, elbow, and ankle. SC of the TMJ is very rare. It is a benign disease that mainly affects unilateral side. It can form cartilaginous and calcified loose bodies of various sizes and cause abnormal function of TMJ.

#### Case Report

In this paper, we report two cases of SC in the upper joint space of the left TMJ. One complained that “Sometimes the left jaw joint feels disoriented” and the other had no symptoms. CT scan and MRI showed left TMJ space widening, multiple tiny calcified mass. After clinical and radiographic analysis, we performed surgical removal of the lesion under general anesthesia. In the histologic examination, synovial chondromatosis was diagnosed in both patients.

#### Conclusions

We report two cases of synovial chondromatosis in the upper joint space of the left TMJ. We performed surgical removal of the lesion. The two patients showed good prognosis without recurrence or pain up to date.

Key words : Synovial chondromatosis, Temporomandibular joint, TMJ

#### Corresponding Author

Chul-Hwan Kim, DDS, MS, PhD

Department of Oral and Maxillofacial Surgery, College of Dentistry, Dankook University

119 Dandae-ro, Dongnam-gu, Cheonan, Chungnam, Korea

TEL : +82-41-550-0271, FAX : +82-41-551-8988, E-mail : kimchoms@dankook.ac.kr

## I . Introduction

Synovial chondromatosis(SC), a proliferative disorder of the synovial membrane was first described by Ambroise Pare in 1558. SC in temporomandibular joint(TMJ) was first reported by Auhausen in 1933<sup>1,2)</sup>. The etiology or cause of SC remains unclear. SC usually occurs in large articular joints. SC of the TMJ is very rare. It is a benign disease that mainly affects unilateral side<sup>3)</sup>. It can form cartilagenous and calcified loose bodies of various sizes and cause abnormal function of TMJ<sup>4)</sup>.

CT and MRI can be used for the diagnosis of SC. After progression of calcification, loose bodies can be identified on CT. However, it may be difficult to identify such loose bodies in many patients. Although MRI is effective in identifying these loose bodies, radiographs and clinical features alone are difficult to diagnose SC accurately<sup>5)</sup>. Therefore, histological examination is essential for accurate diagnosis of SC.

Because nonsurgical procedures are ineffective for SC and loose bodies do not disappear spontaneously, open surgery or arthroscopy with synovectomy are usually performed for SC with surgical removal. Recurrence or malignant transformation after removal of the lesion is known to be rare<sup>6)</sup>.

In this paper, we present two cases of SC in the left temporomandibular joint.

## II . Case Report

### Case 1

A 28-year-old male patient visited the Department of Oral Medicine and Pain in Dankook University Dental Hospital on May 24, 2016. He complained that "Sometimes the left jaw joint feels disoriented". The patient was at the end of orthodontic treatment. The pain and discomfort started in the left TMJ six months ago. There was no other systemic disorder. CT and MRI showed a calcified mass on the left TMJ. The patient was referred to our department of oral and maxillofacial surgery. There was no swelling. However, there was tenderness at the time of palpation and there was pain in the left TMJ at the time of mastication. Mouth opening was over 35 mm. The occlusal relationship was good. There was crepitus sound in the left TMJ at the opening.

CT scan showed left TMJ space widening, multiple tiny calcified mass, and bone erosion (Figure 1). MRI showed left capsule enlargement and multiple low signal intensity in the anterior part of the superior joint space with bone marrow edema in the condyle(Figure 2).

A total of 50-60 loose bodies were removed by exposing the left joint capsule using preauricular incision under general anesthesia(Figure 3).

In the histologic examination, multiple cartilage nodules were seen(Figure 4A). Cartilage nodules were surrounded by a fibrous layer(Figure 4B). Chondrocytes of similar size were uniformly distributed when the inside of the nodule was looked(Figure 4C). Synovial chondromatosis was diagnosed due to the absence of mitosis or

## CASE REPORT

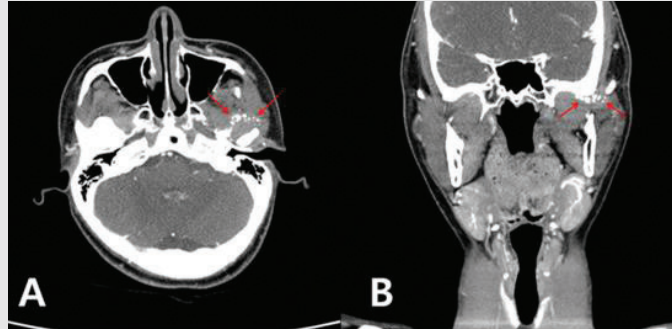


Fig. 1. Enhanced CT image. (A) Axial view. (B) Coronal view. Multiple tiny calcified mass was noted in the left temporomandibular joint (red arrow).

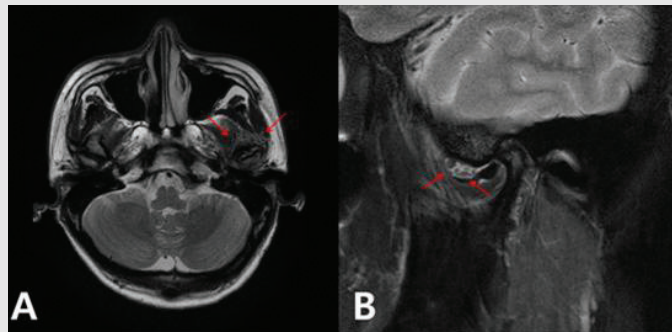


Fig. 2. MRI T2-weighted image. (A) Axial view. (B) Sagittal view. Multiple low signal intensity and effusion was noted in the superior compartment of temporomandibular joint (red arrow).



Fig. 3. 50-60 white loose bodies were removed.

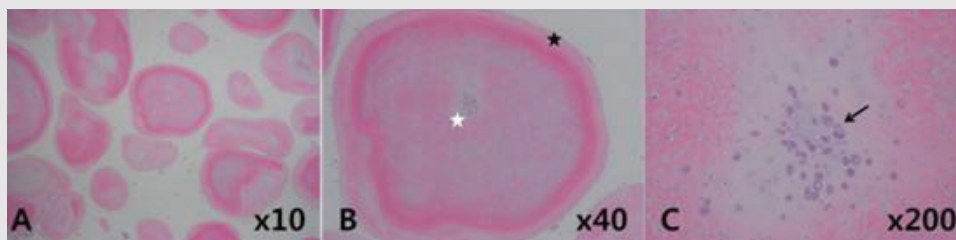


Fig. 4. Histologic features. (A) x10, H&E, Multiple cartilage nodules can be seen. (B) x40, H&E, White stars represent cartilage nodules, which are surrounded by a fibrous layer of black stars. (C) x200, H&E, Chondrocytes of similar size were uniformly distributed when the inside of the nodule was seen.

necrosis suggesting malignancy.

Postoperative CT showed that multiple tiny calcified masses disappeared while the erosion remained(Figure 5). MRI showed that both joint effusion and multiple lesions disappeared after the surgery(Figure 6).

We performed follow-up examinations for 8 months. He showed good prognosis without recurrence or pain.

### Case 2

A 48-year-old man was referred to our clinic on

January 3, 2017 for a radiologically opaque lesions on the left TMJ site in a panoramic view. The patient was treated in various hospitals due to left jaw joint dullness, discomfort, and chewing pain that began 7 years ago. Baseline disease was only hypertension. When the patient was a high school student, he said that he had a pain in the jaw joint and a clicking sound after hit the ball. There was no mouth opening limitation. There was a deviation at the opening toward the affected part. There was no swelling or tenderness. There was no clicking sound either.

CT scan revealed multiple loose bodies

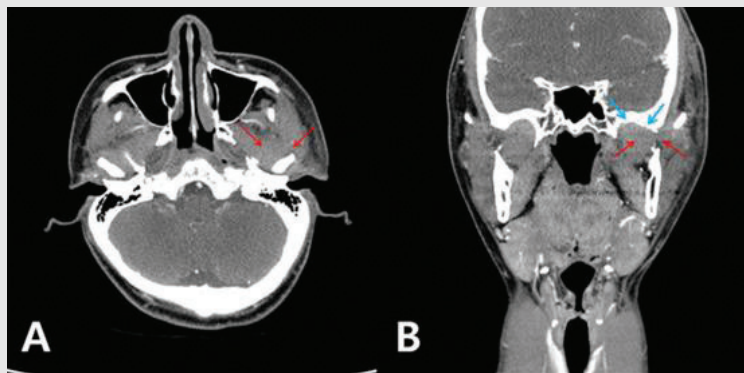


Fig. 5. Postoperative enhanced CT image. (A) Axial view. (B) Coronal view. Multiple tiny calcified masses disappeared(red arrow) while the erosion remained(blue arrow).

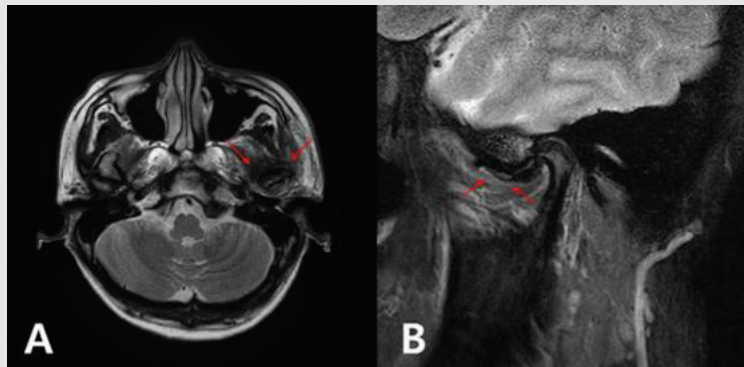


Fig. 6. Postoperative MRI T2-wighted image. (A) Axial view. (B) Sagittal view. Both joint effusion and multiple lesions disappeared after surgery(red arrow).

## CASE REPORT

surrounding the left condyle head(Figure 7). MRI findings showed multiple small low intensity nodular lesions in the affected superior joint space with effusion in the left TMJ(Figure 8).

The left joint capsule was exposed using a preauricular incision to remove three large loose bodies of 1 cm or larger and 100 loose bodies of less than 5 mm(Figure 9).

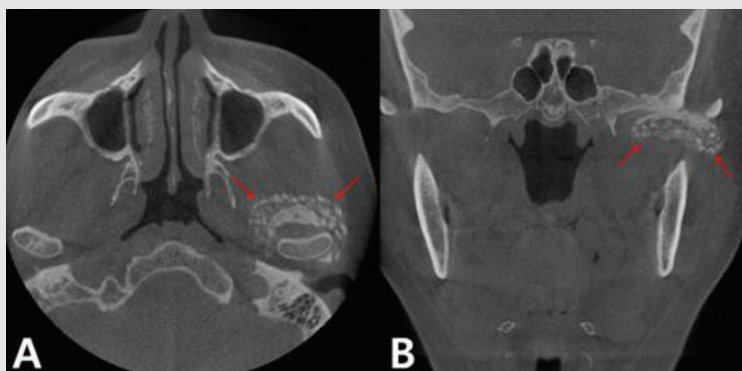


Fig. 7. CBCT image. (A) Axial view. (B) Coronal view. Multiple tiny and several gross calcified mass was noted surrounding the left condyle head (red arrow).

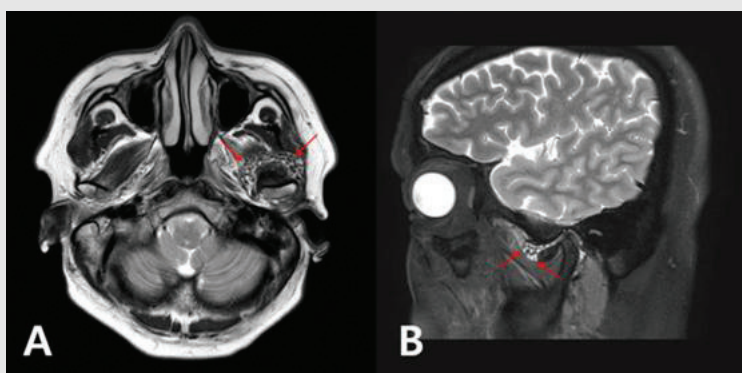


Fig. 8. MRI T2-wighted image. (A) Axial view. (B) Sagittal view. Multiple small low intensity nodular lesions in the affected superior joint space with effusion in left TMJ(red arrow).



Fig. 9. 3 large loose bodies of 1 cm or larger and 100 loose bodies of less than 5 mm were removed.

The histologic examination showed multiple cartilage nodules with a typical SC pattern that a chondrocyte scattered in the nodules and surrounded by a fibrous layer(Figure 10).

The patient showed good prognosis after the operation without any recurrence.

### III. Discussion

The precise etiology of SC and its cause are currently unknown. Although previous trauma, parafunction, degeneration, inflammatory disease, infection, and growth factors have been proposed as contributing factors, there is no accurate evidence to suggest that these are main causes of SC<sup>7</sup>.

Synovial chondromatosis is a proliferative disease of the synovium. It forms a metaplastic cartilaginous nodule on the synovial membrane and pedunculates, eventually falling off into loose body<sup>4</sup>.

SC is mainly involved in large articular joints such as knee, hip, elbow, and ankle. It is more common in men. However, the occurrence of SC

in the TMJ is very rare, with a 1.6: 1 ratio in women/men and a mean age of onset ranging from 40 to 50 ages<sup>4</sup>. TMJ synovial chondromatosis is known to occur in limited upper joint space. Most cases of TMJ SC are unilateral and more common at right TMJ, although bilateral cases are also reported.<sup>3</sup> In the present case report, both patients were males involving the upper joint space of the left TMJ. One patient was in his 20s.

If there is SC in TMJ, various symptoms such as pain, preauricular swelling, mouth opening limitation, clicking, popping, crepitus, malocclusion, vertigo, and tinnitus may occur<sup>8</sup>. However, symptoms of SC in TMJ are not characteristic. In addition, incidence of SC in TMJ is very low. Therefore, it is rare for SC in TMJ to be diagnosed correctly from the beginning. It is often confused with TMJ disorder or parotid tumor<sup>9</sup>.

A metaplastic change in synovium without definite causative factors is called primary SC which is characterized by more aggressive and recurrences. On the other hand, secondary SCs are associated with factors that may stimulate synovium such as previous trauma,

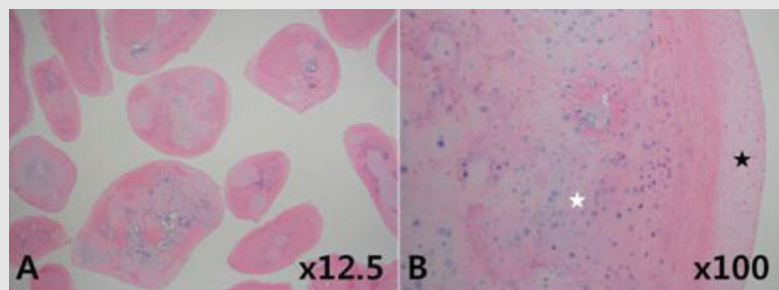


Fig. 10. Histologic features. (A) x12.5, H&E, Multiple cartilage nodules can be seen. (B) x100, H&E, White stars represent cartilage nodules, which are surrounded by a fibrous layer of black stars.

inflammation, arthritis, and so on<sup>9</sup>.

In addition, SC of TMJ can be divided into three stages according to histopathologic features<sup>10</sup>.

1. Initial stage: During this period, metaplasia of the synovial membrane occurs along with proliferation of undifferentiated cells. Detached loose bodies are not yet visible.

2. Transitional stage: It is the time when loose bodies are formed as synovial membrane metaplasia progresses further. Loose bodies containing active chondrocytes are partially surrounded by a synovial membrane.

3. Advanced stage: Detached loose bodies are observed. However, metaplastic activity is not seen on the synovial membrane.

In this case report, the first patient had a history of orthodontic treatment while the second patient had a history of trauma 30 years ago. Although it was suspected to be secondary SC, this history of orthognathic treatment and trauma could not confirm the exact relationship between diagnosis and cause of disease. Both patients underwent histologic examination and found to be in advanced stage of SC.

Conventional X-ray, CT, and MRI can be used for primary diagnosis of SC. Conventional X-rays may be able to identify lesions. However, lesions are less visible when calcification is not fully progressed. Therefore, CT and MRI are most commonly used for diagnosis of SC. Expansion of the joint space and capsule is mostly seen. After calcification, loose bodies can be identified. However, it may be difficult to confirm loose bodies in many patients. MRI is

the best known method for identifying these loose bodies. It has been reported that progressive SC may involve TMJ changes such as bone erosion, sclerosis, disc displacement, and invasion of the middle cranial fossa<sup>5, 11</sup>. For accurate diagnosis of SC, it is necessary to perform a biopsy through arthroscopy or open examination.

Because SC in TMJ is uncommon and its symptoms are similar to normal TMJ disorder, it should be differentiated from secondary synovial chondrometaplasia due to degenerative, inflammatory, metabolic, and traumatic joint disease. It also should be differentiated from condylar hyperplasia and cartilaginous neoplasms<sup>12</sup>.

Differential diagnosis of SC with chondrosarcoma is needed. Chondrosarcoma in TMJ is extremely rare. Up to date, only 20 such cases have been reported<sup>13</sup>. Condyle, temporal bone, and synovial membrane are sites of chondrosarcoma development. It is prevalent in women in their 40s. TMJ disorder symptoms can be noted in chondrosarcoma. Radiographs may show expansible mass involving TMJ, condyle, and infratemporal fossa<sup>13</sup>.

Histologic findings of SC can be seen in various sizes of cartilaginous cellular nodules. The border of the nodule is composed of hyaline fibrocartilaginous tissue. There may be synovial lining depending on progression of the lesion.<sup>14</sup> In this cases, both histologic examinations showed multiple cartilage nodules with a typical SC pattern that a chondrocyte scattered in the nodules and surrounded by a fibrous layer.

Because SC does not disappear spontaneously or respond to non-surgical procedures, treatment usually involves surgical removal of the lesion. Surgical procedure depends on the size of the nodule, the anatomical structure involved, and the degree of progression of the lesion. The most commonly used treatment is the removal of cartilaginous bodies by an open surgery with synovectomy for the lesion.<sup>6</sup> Arthroscopic removal of the lesion is a successful treatment when the lesion is confined to a single joint compartment, when extra-articular extension is not seen<sup>15</sup>.

Recurrence after removal of the lesion is very rare. Several review papers have confirmed that synovectomy is not performed in recurrent cases<sup>16</sup>. Therefore, removal of the affected synovium is necessary.

#### IV. Conclusions

In summary, we report two cases of synovial chondromatosis in the upper joint space of the left TMJ. We performed surgical removal of the lesion. The two patients showed good prognosis without recurrence or pain up to date.

#### V. Acknowledgement

The authors claim to have no financial interests, either directly or indirectly, in the products or information listed in this article.

Written informed consent was obtained from the patient for the publication of this report and any accompanying images.

## 참 고 문 헌

1. Koyama J., Ito J., Hayashi T., and Kobayashi F., Synovial chondromatosis in the temporomandibular joint complicated by displacement and calcification of the articular disk: report of two cases. *AJNR Am J Neuroradiol*, 2001. 22(6): p. 1203-6.
2. Wong W.C., Cheng P.W., and Chan F.L., MRI appearance of synovial chondromatosis in the temporomandibular joint. *Clin Radiol*, 2001. 56(9): p. 773-4.
3. Guijarro-Martinez R., Puche Torres M., Marques Mateo M., Solis Garcia I., Miragall Alba L., Iglesias Gimilio M.E., Perez-Herrezuelo Hermosa G., and Pascual Gil J.V., Bilateral synovial chondromatosis of the temporomandibular joint. *J Craniomaxillofac Surg*, 2011. 39(4): p. 261-5.
4. Testaverde L., Perrone A., Caporali L., Ermini A., Izzo L., D'Angeli I., Impara L., Mazza D., Izzo P., and Marini M., CT and MR findings in synovial chondromatosis of the temporo-mandibular joint: our experience and review of literature. *Eur J Radiol*, 2011. 78(3): p. 414-8.
5. Kahraman A.S., Kahraman B., Dogan M., Firat C., Samdanci E., and Celik T., Synovial chondromatosis of the temporomandibular joint: radiologic and histopathologic findings. *J Craniofac Surg*, 2012. 23(4): p. 1211-3.
6. Chen M.J., Yang C., Cai X.Y., Jiang B., Qiu Y.T., and Zhang X.H., Synovial chondromatosis in the inferior compartment of the temporomandibular joint: different stages with different treatments. *J Oral Maxillofac Surg*, 2012. 70(1): p. e32-8.
7. Martin-Granizo R., Sanchez J.J., Jorquera M., and Ortega L., Synovial chondromatosis of the temporomandibular joint: a clinical, radiological and histological study. *Med Oral Patol Oral Cir Bucal*, 2005. 10(3): p. 272-6.
8. Meng J., Guo C., Yi B., Zhao Y., Luo H., and Ma X., Clinical and radiologic findings of synovial chondromatosis affecting the temporomandibular joint. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*, 2010. 109(3): p. 441-8.
9. Zha W., Zhao Y.F., Liu Y., and Jiang L., A case of synovial chondromatosis of the temporomandibular joint secondary to preauricular trauma. *Int J Oral Maxillofac Surg*, 2009. 38(11): p. 1212-5.
10. Milgram J.W., The classification of loose bodies in human joints. *Clin Orthop Relat Res*, 1977(124): p. 282-91.
11. Xu W.H., Ma X.C., Guo C.B., Yi B., and Bao S.D., Synovial chondromatosis of the temporomandibular joint with middle cranial fossa extension. *Int J Oral Maxillofac Surg*, 2007. 36(7): p. 652-5.
12. Sink J., Bell B., and Mesa H., Synovial chondromatosis of the temporomandibular joint: clinical, cytologic, histologic, radiologic, therapeutic aspects, and differential diagnosis of an uncommon lesion. *Oral Surg Oral Med Oral Pathol Oral Radiol*, 2014. 117(3): p. e269-74.
13. Gonzalez-Perez L.M., Sanchez-Gallego F., Perez-Ceballos J.L., and Lopez-Vaquero D., Temporomandibular joint chondrosarcoma: Case report. *J Craniomaxillofac Surg*, 2011. 39(1): p. 79-83.
14. Davis R.I., Hamilton A., and Biggart J.D., Primary synovial chondromatosis: a clinicopathologic review and assessment of malignant potential. *Hum Pathol*, 1998. 29(7): p. 683-8.
15. Cai X.Y., Yang C., Chen M.J., Jiang B., Zhou Q., Jin J.M., Yun B., and Chen Z.Z., Arthroscopic management for synovial chondromatosis of the temporomandibular joint: a retrospective review of 33 cases. *J Oral Maxillofac Surg*, 2012. 70(9): p. 2106-13.
16. D'Souza B. and Dimitroulis G., A case of recurrence of synovial chondromatosis of the temporomandibular joint. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*, 2007. 104(4): p. e59-61.