

Synovial Chondromatosis of the Temporomandibular Joint: A Case Report

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Synovial chondromatosis (SC) is a rare, benign disease found in metaplastic cartilaginous nodules within the synovial membrane. The etiology of SC is not well-known, but it may be associated with trauma, chronic abnormal loading, or inflammatory joint disease. It is often found in knee, elbow, ankle, and shoulder joint but rarely in the temporomandibular joint (TMJ) area. SC of TMJ frequently appears in middle age and more often in females. Clinical symptoms include periarticular swelling, pain, crepitus, and limitation of joint motion. The most common feature is a radiographic finding of loose bodies in the joint. Irregularity of joint space and condylar head is also observed. Treatment involves the surgical removal of synovium and loose bodies. The prognosis is good, with low postsurgical recurrence rate. Functional improvement of the joint and pain relief are noted in many patients. Malignant transformation of SC has not been recorded. In this study, we report a 54-year-old female patient who experienced discomfort on the right TMJ with SC on the area but made a satisfactory recovery after surgery.

Key Words: Chondromatosis, synovial; Temporomandibular joint

Introduction

As a comparatively rare disease wherein cartilaginous nodules are formed in the synovial membrane including loose bodies within the articular cavity, synovial chondromatosis (SC) may be defined as the cartilaginous denaturalization of the synovial membrane¹⁻³. It is said to be

observed frequently in the cartilaginous area of large joints such as knee and elbow, with more than 50% of the disease occurring in knee joints. It is a benign disease that is rarely found in the temporomandibular joint (TMJ)^{2,4}. Causes of the disease are not clearly known, but they are believed to include genetic cause, regressive articular disease, continued stimulation of the dislocated articular

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disk, trauma, and inflammation.

SC of the TMJ frequently affects middle-aged females, and edema, pain, joint noise, and mouth-opening limitations are observed in the contracted area. When the mouth is opened, various symptoms including declination toward the contracted area or recurrent dislocation of the TMJ--similar to the symptoms of normal temporomandibular joint disturbance--may occur. If the symptoms are serious, malocclusion carrying cross-bite may occur. Radiologically, sometimes, no special variations are noted in the joint area, but nodules of radiopacity in areas surrounding the glenoid cavity can be identified, and most of them exist in the articular cavity. Sometimes, some of them come out from the glenoid cavity and invade the cranial

cavity. Articular regression is sometimes found, but not bone destruction such as malignant tumor in general. Even though treatment as a general rule involves the surgical removal of the nodules and the contracted synovial membrane, articular discectomy, TMJ reconstruction, and condylectomy



Fig. 1. The panoramic view on the first visit shows numerous masses around the right condylar process with bone density.



Fig. 2. (A~C) The preoperative facial computed tomography showed calcified loose bodies at the lateral and medial parts of the right condyle. (D) The right condylar head had osteophytic change.

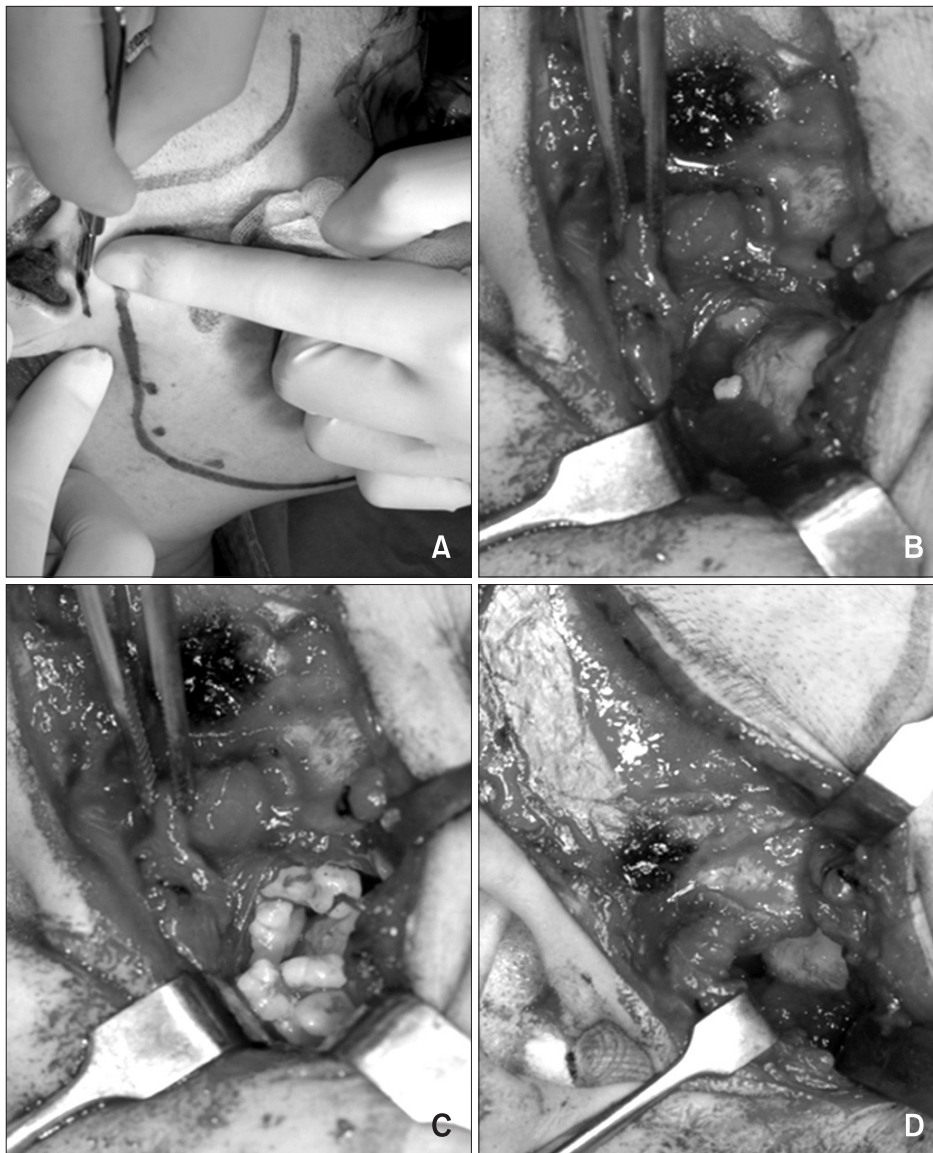


Fig. 3. (A~D) Preauricular incision and intraoperative retrieval of multiple cartilaginous masses.

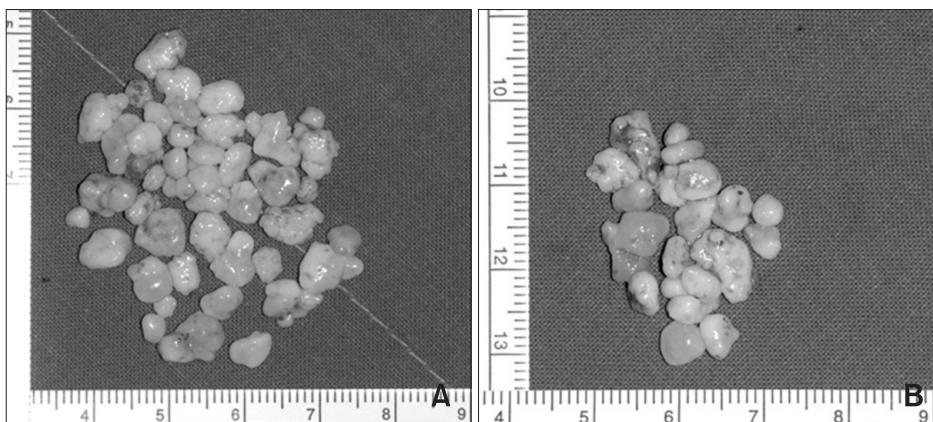


Fig. 4. Numerous loose bodies whose diameter ranged from 0.5 mm to 8.0 mm and numbering about 50 and 20 were removed from the medial (A) and lateral (B) side condylar head, respectively.

may also be performed depending on the severity of the disease.

The case, after surgery, is somewhat less likely to lead to recurrence and change to malignant state. This case involves a 50-year-old female patient who came to the hospital complaining of discomfort of

the right TMJ during mouth opening or closing. The patient was diagnosed with SC based on clinical and X-ray tests. Since the patient showed favorable results after surgical treatment, this paper reports the case along with a review of the relevant literature.

Case Report

A female patient in 50s visited the hospital complaining of discomfort of the right-side TMJ area when opening or closing her mouth, which started 7~8 years ago. Clinical trials showed minor edema on the right-side TMJ, and she complained of oppressive pain when tactile diagnosis was conducted. There was a slight sense of stiffness and solidification, but no wave was felt. Maximum



Fig. 5. Postoperative panorama: no remnants around the right condylar process after operation.

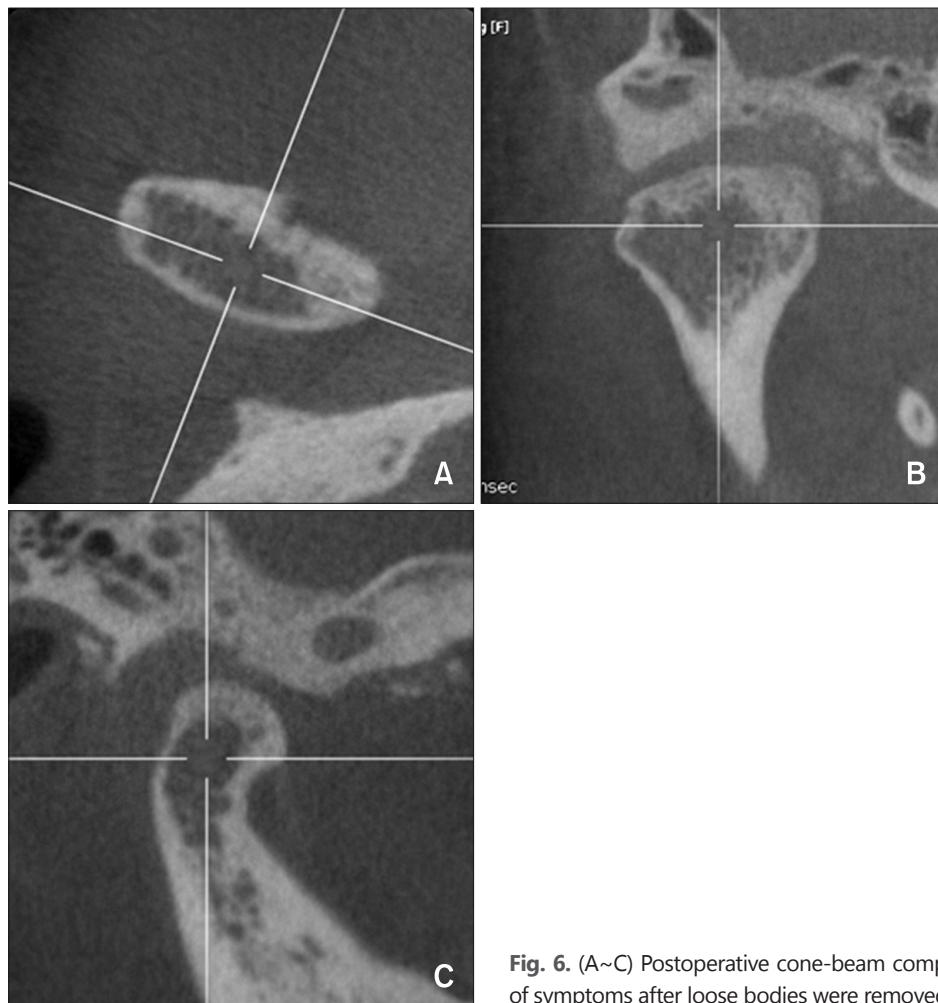


Fig. 6. (A~C) Postoperative cone-beam computed to-mography: improvement of symptoms after loose bodies were removed.

mouth opening was approximately 45 mm, indicating no mouth-opening disorder, but there was joint noise from the right side, and clicking sound was intermittently heard. There was no declination of the lower jaw during mouth opening or closing, and the cross-bite of the upper and lower jaws was comparatively stable. Neither was there special systemic disease or abnormal oral habit of the patient or medical history of trauma on joints.

During the initial diagnosis, the panorama photo showed many radiopaque materials in the vicinity of the right-side condylar (Fig. 1). The computed tomography photo showed the articular cavity of the right-side TMJ, which has been somewhat widened, and many calcified nodules were observed on the right and left sides of the soft tissue of the condylar. In addition, aspects of regressive diseases such as flattening of the right-side joint cavity and the condylar and irregularity of the border and hardening phenomenon were observed,

albeit minor, and the right masseter was deflected toward the inside by the disease (Fig. 2).

The case was diagnosed as SC after consolidating the aforesaid findings, and surgically removing the nodules was planned. The patient was placed under general anesthesia, and access was made to the right TMJ using the preauricular approach. The lateral capsule was in an expanded state, and during the process of removing the lateral capsule, a large amount of pericardial effusion with color similar to rice straw flowed out; there were also many loose bodies discovered outside the right condylar and the glenoid cavity, and they were removed (Fig. 3). A visual inspection confirmed that there was no morphologic disorder on the glenoid cavity and the condylar, and they were delaminated toward inside the joint. After verifying the existence of numerous loose bodies that used to be located inside the joint, they were removed without additional oral excision. Since the range of the disease did not

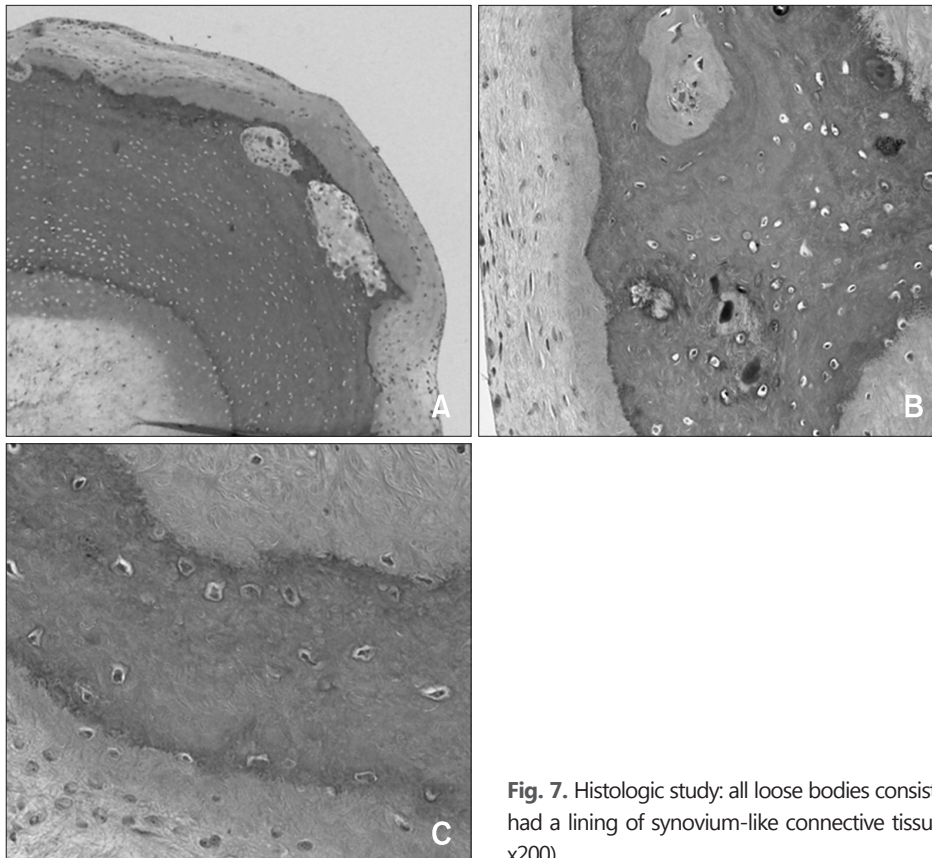


Fig. 7. Histologic study: all loose bodies consist of mature cartilage, some of which had a lining of synovium-like connective tissue (H&E staining; A, x40; B, x100; C, x200).

intrude other anatomical structures, and there were no anatomical morphologic disorder noted in the vicinity of the joint, no additional surgical operation was performed. The removed cartilaginous loose bodies had a diameter of 0.5 mm in the case of the smallest one and 8.0 mm in the case of the largest one, which was white and firm. The surface created in the form of rice grain was clean, and some were

lumped together. A total of 50 loose bodies were removed from inside the joint, and 20 loose bodies, from the outside (Fig. 4).

After surgical operation, the loose bodies that used to exist prior to the surgical operation were verified to have been removed completely through the panorama and cone-beam computed tomography (CBCT) image (Figs. 5, 6). Immediately after the operation, dropping phenomenon of the right eyebrow and mouth-opening limitations were observed, but they did not persist when checked after 2 weeks. Pathological tests were conducted on the removed loose bodies, with the results revealing mature cartilaginous tissue surrounded by connective tissue similar to the synovial membrane (Fig. 7). For up to 10 months after surgery, there were no symptoms of recurrence. The regressive symptoms of the right-side joint were still observed



Fig. 8. Postoperative panorama, follow-up 10 months.

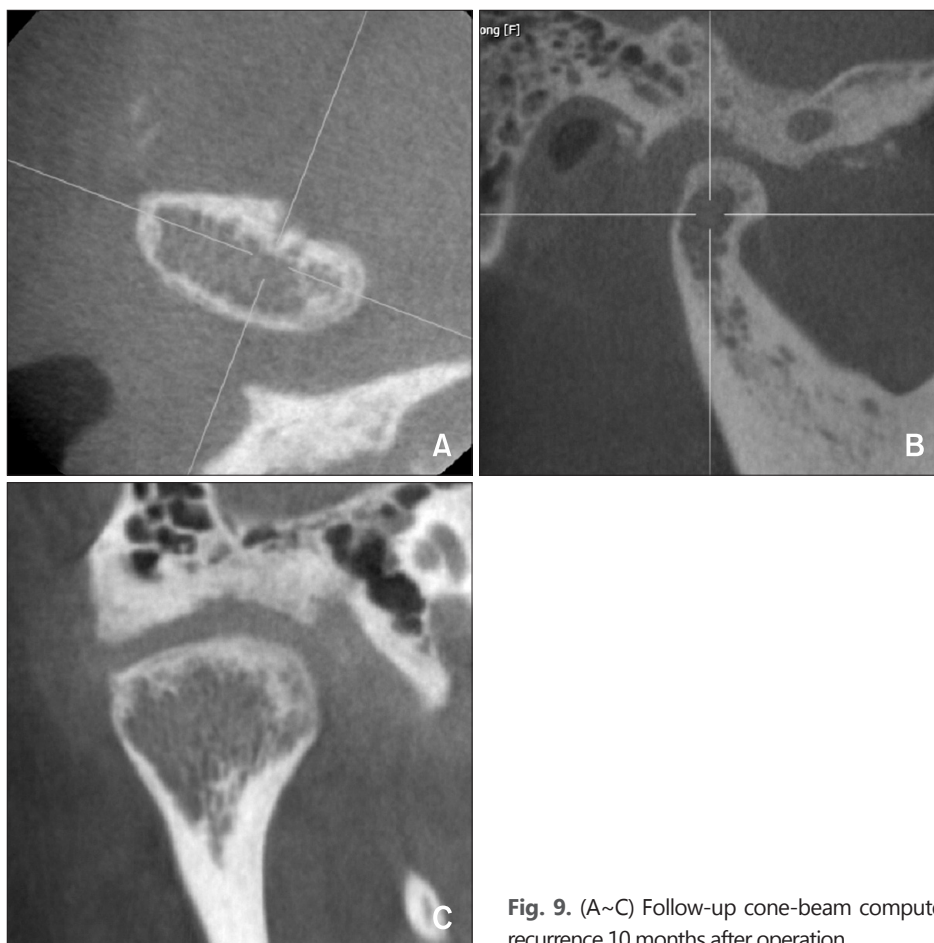


Fig. 9. (A~C) Follow-up cone-beam computed tomography study showing no recurrence 10 months after operation.

through the CBCT image compared with the prior symptoms, but there was neither aggravation nor clinical discomfort on the part of the patient (Figs. 8, 9).

Discussion

Even though the causes of SC are unclear, some of them are believed to include trauma, inflammation, and cartilage debris and are regarded as the process of the positive tumors of the synovial membrane or denaturalization³⁾. The disease may develop due to regressive joint disease or abnormal pressure resulting from malocclusion or displaced articular disease, and this was reportedly caused by metaplasia, not neoplasia⁵⁾. The cartilaginous cell of loose bodies was derived from the mesenchymal cell inside the synovial membrane, and denaturalization may be induced by trauma or inflammation. It is called primary SC if the causes of disease are unclear or secondary SC if there are related factors such as trauma or inflammation. In the case of primary SC, it develops less frequently but is said to be more aggressive, showing a high recurrence rate after surgery^{1,6-9)}. The SC that has developed from joints other than the TMJ develops more frequently in males in general. When the disease occurs on the TMJ, however, its frequency is higher in females (7 : 3). This is believed to be due to the fact that TMJ disease occurs more frequently in females. It develops more frequently on the right side (1.25 : 1), and sometimes, although rare, occurs on both sides⁸⁻¹⁰⁾.

Clinical symptoms show various aspects such as swelling of the TMJ, limitations of mandibular movement, pains when tactile diagnosis is carried out, and joint noise; in some cases, they are accompanied by changes in cross-bite. Since these are similar to the symptoms of inflammation on the TMJ and regressive disease, it is sometimes difficult to diagnose the case directly as SC if there are no special radiological findings^{8,11)}. In this case, the patient came to the hospital chiefly complaining of

discomfort on the right-side TMJ, which persisted for several years. Even though there were no mouth opening limitations, joint noise, sense of stiffness on the right-side TMJ, oppressive pain in the adjacent area when tactile diagnosis was conducted, and minor case of edema were observed, and they were similar to the symptoms exhibited by patients suffering from ordinary TMJ diseases.

The radiological findings of SC varied depending on the level of calcification or ossification; sometimes, radiopaque lumps are shown inside the synovial membrane, or many loose bodies are observed inside the TMJ. If the loose bodies are not calcified, they cannot be observed from ordinary X-ray photos. In some cases, regressive changes such as erosion of the increased articular cavity, glenoid cavity, or condylar may occur. As for the range of disease invasion, it is limited to inside the joint in most cases. There are cases wherein the disease expands to the outside of the articular capsule^{2,3,8)}. According to reports, the disease may sometimes invade the inside of the forward soft tissue of the cranial cavity, external auditory meatus, parotid gland, or articular process; it may be accompanied by neurologic symptoms like facial paralysis if it develops inside the cranial cavity⁸⁾. In this case, it was verified through the CT image that the right articular capsule has been expanded, and that many masses have invaded the soft tissue surrounding the joint. Since they were located in 2 places, i.e., internal and external sides of the joint, the possibility of performing surgical excision if necessary--by making additional oral access--was considered. The inside articular capsule was expanded considerably by pericardial effusion, and this seems to have been caused by the edema and discomfort felt prior to surgery. The existence of loose bodies around the TMJ is the most characteristic symptom of SC; such may also be observed in osteochondritis dissecans, regressive TMJ disease, rheumatoid arthritis, chondrosarcoma, or chronic renal failure, but rarely⁸⁾. In the case of

SC, the number of loose bodies having diameter of 0.5~8.0 mm was 50 and 20 from the internal and external sides, respectively.

As the pathological characteristics of SC, in general, many cartilaginous nodes are generated on one synovial membrane. The cartilaginous nodes existing in the synovial membrane may be located loosely in the articular cavity and may often be calcified or ossified. Histologically, such nodes and loose bodies show a hyaline cartilage aspect and the appearance of partially progressed ossification. Often, moderate cellular atypia and dikaryotic cells are observed. Cartilages display hyperchromatism and, sometimes, an atypical appearance carrying cartilaginous cell featuring 2 nuclei; in such case, differential diagnosis is needed with cartilaginous sarcoma⁹⁾. Milgram histologically classified SC into 3 stages; the early stage is the dysplasia of the synovial membrane cell not carrying cartilaginous loose bodies. The transitional stage is the dysplasia of the loose bodies and synovial cell, and the advanced stage is a case wherein there is no dysplasia of the synovial membrane, and only node loose bodies--ossified in various sizes--exist⁸⁾. The method of treatment may vary according to such histologic stages, and more aggressive approach is needed in the case of the early and transitional stages^{3,12)}. In case symptoms of SC are discovered, the treatment principle is to remove the loose bodies existing in the joint capsule. If only regressive changes are shown radiologically without loose bodies, however, preservative therapy including physical therapy, steroid injection, or occlusal adjustment is sometimes proposed; if necessary, aspiration designed to remove the pericardial effusion or method of injecting hyaluronidase in the joint may be considered¹³⁾. If loose cartilage is created, however, the disease does not auto-genously react to preservative therapy. In such case, a more appropriate therapy would be to remove the denaturalized cartilaginous disease and the synovial membrane⁴⁾. In case the disease progresses

to a serious level, articular disc removal may be performed if the disc is perforated; if masses are located inside the condyle, making it difficult to remove the masses completely without removing the condylar, condylectomy may be considered¹³⁾.

If the number of loose bodies is small, they exist only in the upper articular cavity, and their sizes are less than 3 mm, removing loose bodies through arthroscopy may also be considered.

In particular, if the disease cannot be diagnosed radiologically, it is reportedly possible to diagnose and remove masses through the use of arthroscopy and to expect good results while minimizing patients' discomfort using a less invasive method^{2,5,10,14)}. It is known that, even though prognosis after the surgery is good, the remaining synovial membrane may lead to the denaturation of condyle cartilage; in case of remaining cartilaginous disease or overproliferated synovial membrane, the disease may recur, but the likelihood of such is comparatively low^{10,12,15)}. Even though there are no causes of recurrence disclosed, long-term follow-up study after surgery is required. Moreover, malignant translocation of this disease has yet to be reported.

In this case, the patients' symptoms were not serious, foregoing the need to invade the skull or mandibular condyle; only the created loose bodies were removed to preserve the anatomical structure and functions. There were no symptoms of recurrence or malignant translocation discovered for up to 10 months after surgery, and normal exercise functions were done without discomfort.

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