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# Giant Cell Lesions of the Temporomandibular Joint Area: A Report of Three Cases

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## ABSTRACT

Giant cell lesions include central giant cell granuloma and tenosynovial giant cell tumor. Central giant cell granuloma is a rare benign giant cell lesion formerly classified as a subtype of a giant cell tumor of the bone. In cases of central giant cell granuloma in the temporomandibular joint, the growth of the lesion may destroy nearby anatomic structures like the infratemporal fossa or mandibular condyle. Surgical removal of the lesion is required; however, only curettage increases the possibility of recurrence. Tenosynovial giant cell tumors are benign tumors arising from tendon sheaths of various joints. They commonly affect the joints of long bones, but some cases of tenosynovial giant cell tumors in the temporomandibular joint area have been reported. This case report is based on three patients diagnosed with giant cell lesions in the temporomandibular joint who underwent surgical treatment. Through this case report and a review of the relevant literature, we discuss the diagnosis and treatment of giant cell lesions in the temporomandibular area and report cases of immediate reconstruction using an alloplastic total joint reconstruction prosthesis.

**Key words :** Central giant cell granuloma, Tenosynovial giant cell tumor, Pigmented villonodular synovitis, Temporomandibular joint, Total joint replacement

## INTRODUCTION

Different types of multinucleated giant cells have been identified in oral biopsies. For example, macrophage fusion can form giant cells under granulomatous inflammation like tuberculosis. In some cases, reactions with keratin or fat crystals can make giant cells[1]. There have been numerous attempts to classify oral lesions with giant cells. Rosenberg et al.[2] described giant cell granuloma and pigmented villonodular synovitis as benign giant cell lesions of the bone.

Central giant cell granuloma (CGCG) is a rare benign lesion of the jaw bone that was first distinguished from giant cell tumors (GCT) of the bone by Jaffe[3]. Since Jaffe's report, many researchers have tried to compare CGCG and GCT to help in differential diagnoses. According to Auclair et al.[4], the most commonly affected primary location of GCT is the femur and tibia, while CGCG often occurs in the mandible and maxilla. This research suggested that GCT and CGCG share a single disease process but differ from the site of occurrence and the age of patients. However, Nagar et al.[5], in 2020, reported that p63 immunohistochemical

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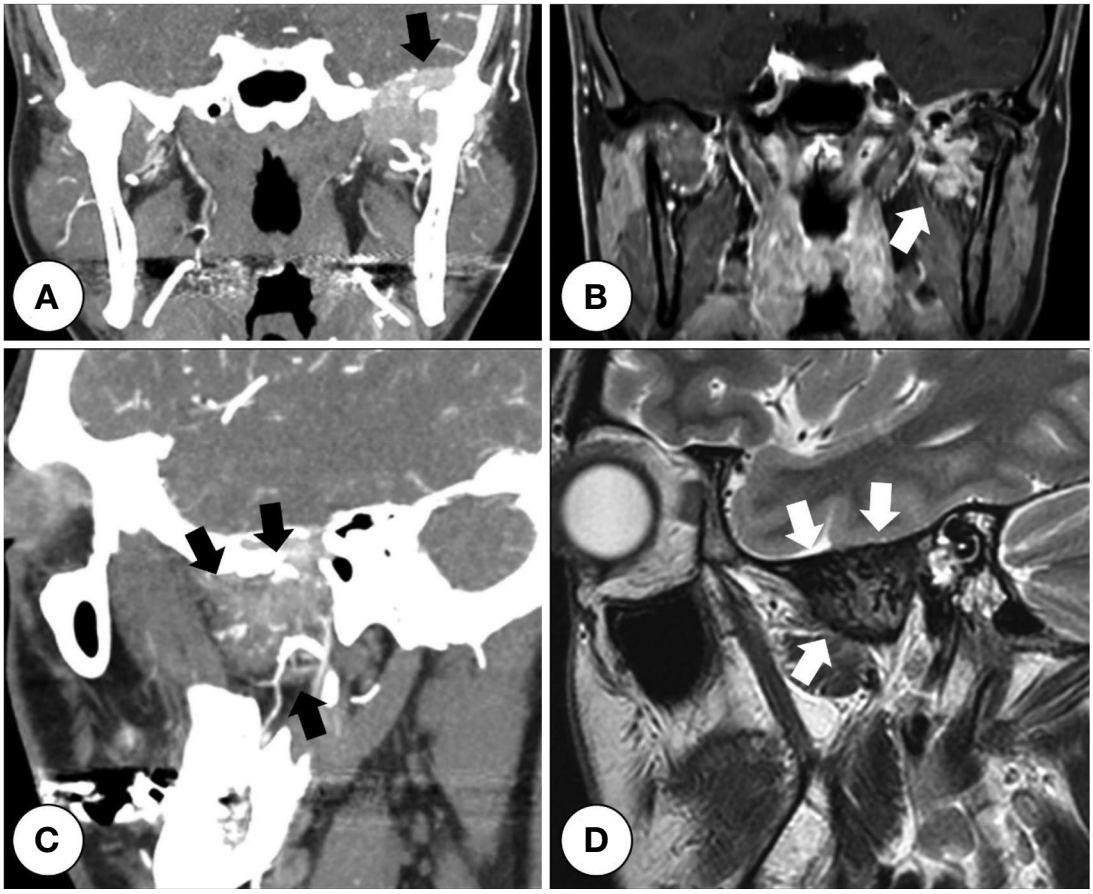


***Figure 1.*** *Imaging features of central giant cell granuloma (Case 1). A. Coronal view of contrast-enhanced computed tomography (CT) image presents a well-encapsulated, lobulated enhancing mass with bony destruction of condylar head and temporal bone (black arrow). B. On gadolinium-enhanced T1-weighted magnetic resonance image (MRI), lesion reveals enhancement with internal dark foci on medial side of the mandibular condyle (white arrow). C. Sagittal view of contrast-enhanced CT presents the mass described above at the anterior and inferior of the articular eminence (black arrow). D. Sagittal view of T2 weighted MR image shows slightly high signal intensity surrounded by dark signaled rimming suggesting the hemosiderin deposit (white arrow).*

staining can differentially diagnose CGCG and GCT. While 100% of GCT cases showed p63 immunoexpression, none of the CGCG cases showed p63 immunopositivity. With this report, GCT and CGCG can be differentially diagnosed and divided into completely different entities. The incidence of CGCG in the craniofacial area was measured as 0.00011% in Netherlands from 1990 to 1995, and only five cases of CGCG in the mandibular condyle were reported until 2020[6].

Tenosynovial giant cell tumors (TSGCT), also known as pigmented villonodular synovitis are rare benign neoplasms that occur in the tendon sheath or synovium of various joints. TSGCT usually affects the knee, hip or shoulder; however, few cases of temporomandibular joint (TMJ) have been reported[7]. The clinical symptoms of TSGCT are pain and swelling in the affected joint area and limitation of joint movement. In normal cases, symptoms gradually worsen and are accompanied by joint damage and degenerative changes, resulting in severe functional impairment of the joint[8]. When TSGCT occurs in the TMJ area, the patient complains of limited mouth opening and pain and swelling in the preauricular region. It is sometimes observed in the form of a soft tissue mass on radiographic images; however, it is difficult to differentiate without magnetic resonance imaging (MRI) when there is no damage to adjacent bone or cartilage tissue. Therefore, total surgical resection of the lesion is strongly recommended[9].

In this paper, we report the cases of three patients diagnosed with giant cell lesions (GCL) of the TMJ area (one CGCG, one

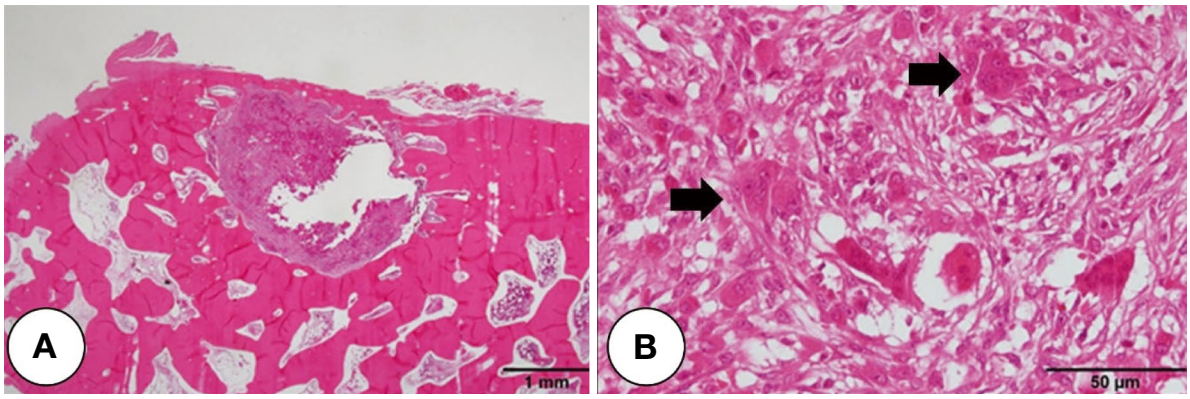


***Figure 2.*** *Histopathologic examination (Case 1). A. B. Microphotography after Hematoxylin and eosin staining. Resected condylar head revealed nodular mass destroying condylar medulla and cortex bone. The mass consisted of numerous osteoclast-like giant cells, mononuclear cells, which were both oval and spindle in shape, and eosinophilic cytoplasm, hemorrhage, and vascular stroma (A: ×12.5 magnification, B: ×400).*

TSGCT, and one recurrent TSGCT) who underwent surgical treatment along with a literature review. In two cases, because the resection range was wide, total joint replacement (TJR) using alloplastic prostheses of Biomet® (Zimmer Biomet, Warsaw, IN, USA) was immediately performed. In cases with a lower resection range, only the mass excision showed good recovery without any remarkable recurrence. We would like to share opinions on the diagnosis, surgery, and rehabilitation of GCLs in the TMJ through case reports of three patients successfully treated without major postoperative complications.

## CASE REPORTS

### Case 1

A 40-year-old female patient was referred from the Department of Neurosurgery for the co-operation of the left TMJ area. The patient complained of persistent tinnitus and discomfort in the left ear. Through radiographic examinations at another hospital's department of otolaryngology, GCT of the temporal area was suspected. The maximum incisor opening (MIO) was 37 mm. The patient complained of stinging discomfort in the left TMJ at the time of maximum opening, and deflection to the left was observed during mouth opening.

In the panoramic and TMJ panoramic views, bone resorption of the left anterior condyle was observed. An external computed tomography (CT) image showed an osteolytic lesion on the medial side of the left temporal bone and mandibular condyle, and an MRI image showed a 2.4×2.3 cm mass in the same area. (Fig 1A-D)

Surgical resection of the patient's mass and reconstruction of the mandibular condyle using alloplastic total joint replacement system of Biomet® were planned. The operation was performed in collaboration with otolaryngology, neurosurgery, and oral and maxillofacial surgery departments under general anesthesia. The mass was exposed using a retroauricular approach, and the medial side of the mandibular condyle showed severe bone resorption due to the mass. Unlike the cases of other TJRs, in this patient, the fossa component could not be positioned in the articular fossa due to the resorption of the cranial base; therefore, it had to be fixed at the zygomatic arch.

The resected specimen was diagnosed as CGCG based on histopathological examination (Fig 2A, B). Self-opening exercise was provided for the patient's rehabilitation after surgery, and MIO was evaluated at each follow-up visit. After surgery, there were no postoperative complications such as facial nerve damage or Frey's syndrome. It has been approximately 3 years since the surgery, and follow-up CT, MRI examinations were performed at every visit. Follow-up images showed no recurrence of the tumor and no complications of alloplastic joint implants, such as infection or screw loosening. The patient showed no discomfort in mastication and pronunciation and good recovery with an MIO of 44 mm without pain.

### Case 2

A 33-year-old male patient visited our department complaining of pain in the right TMJ area that had occurred for

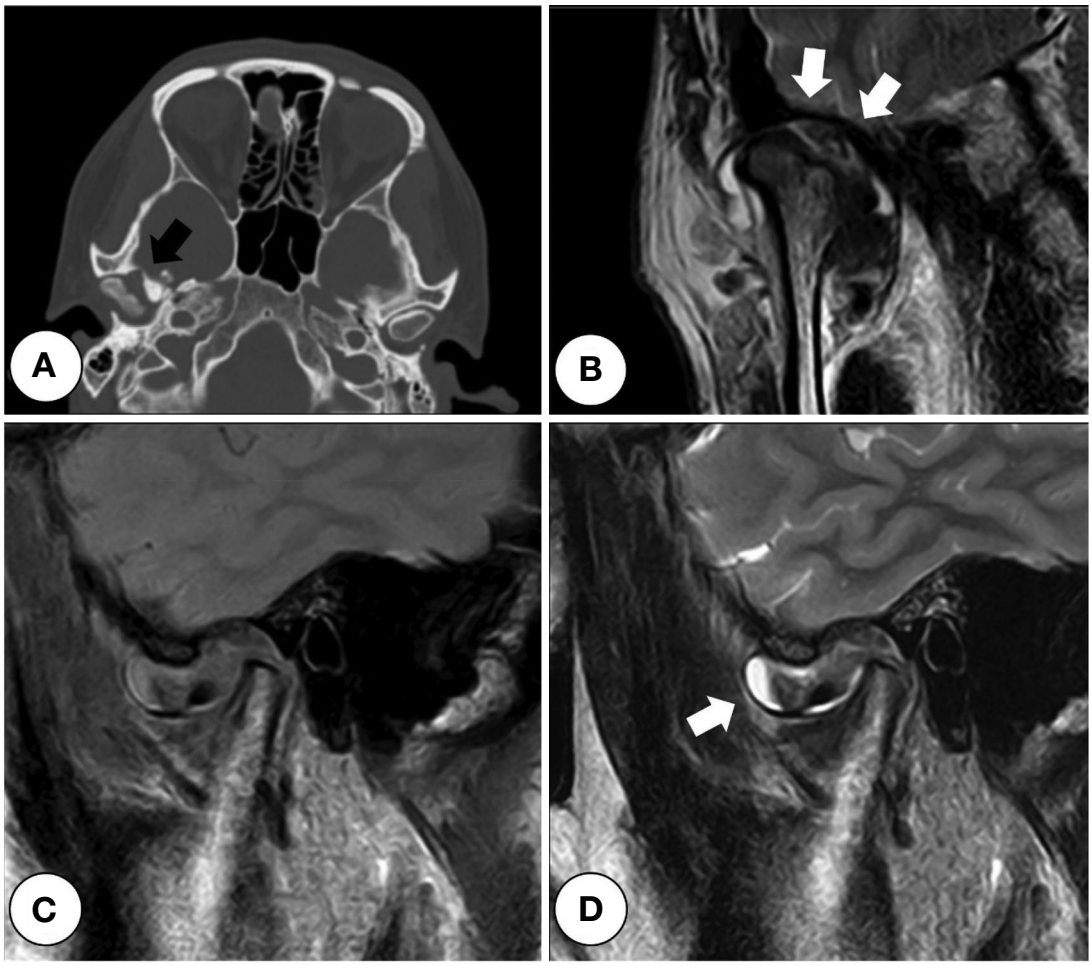


***Figure 3.*** *Imaging features of tenosynovial giant cell tumor (Case 2). A. Axial bone setting CT image shows compressive remodeling on the anterior slope of the articular fossa (black arrow). B. Coronal view of T2 weighted MR image presents low signal intensity (SI) with internal patchy-like heterogeneity on the right TMJ (white arrow). C, D. Sagittal MR images show anteriorly displaced disc and low SI tumor mass filling the superior joint space on T1 and T2 weighted images. Joint fluid collection (white arrow) is observed, which could suggest an inflammatory process regarding the etiology of the giant cell lesion.*

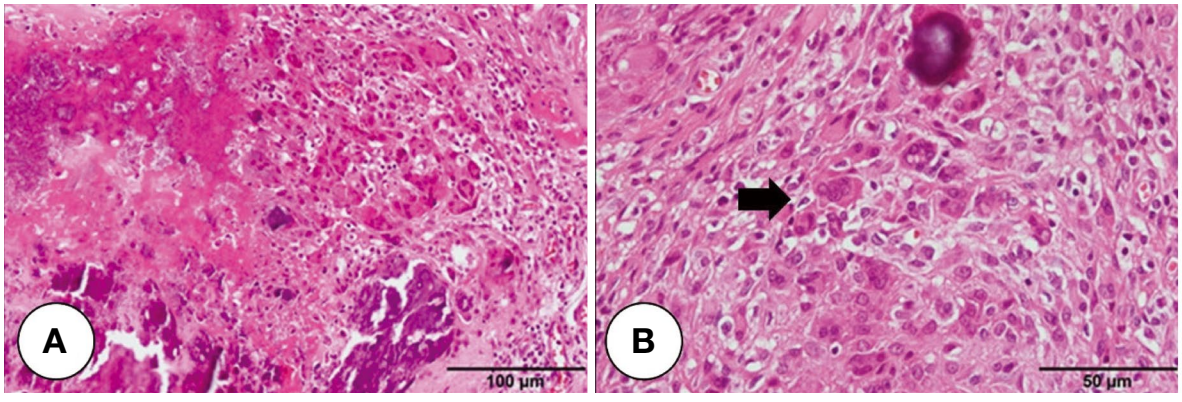


***Figure 4.*** *Histopathologic examination (Case 2). A. B. Microphotography after Hematoxylin and eosin staining. The tumor consisted of multinucleated giant cells, histiocytic mononuclear cells, and extravasated red blood cells. Also, prominent chondroid metaplasia was observed in the geographic pattern within the tumor (A: ×200, B: ×400).*

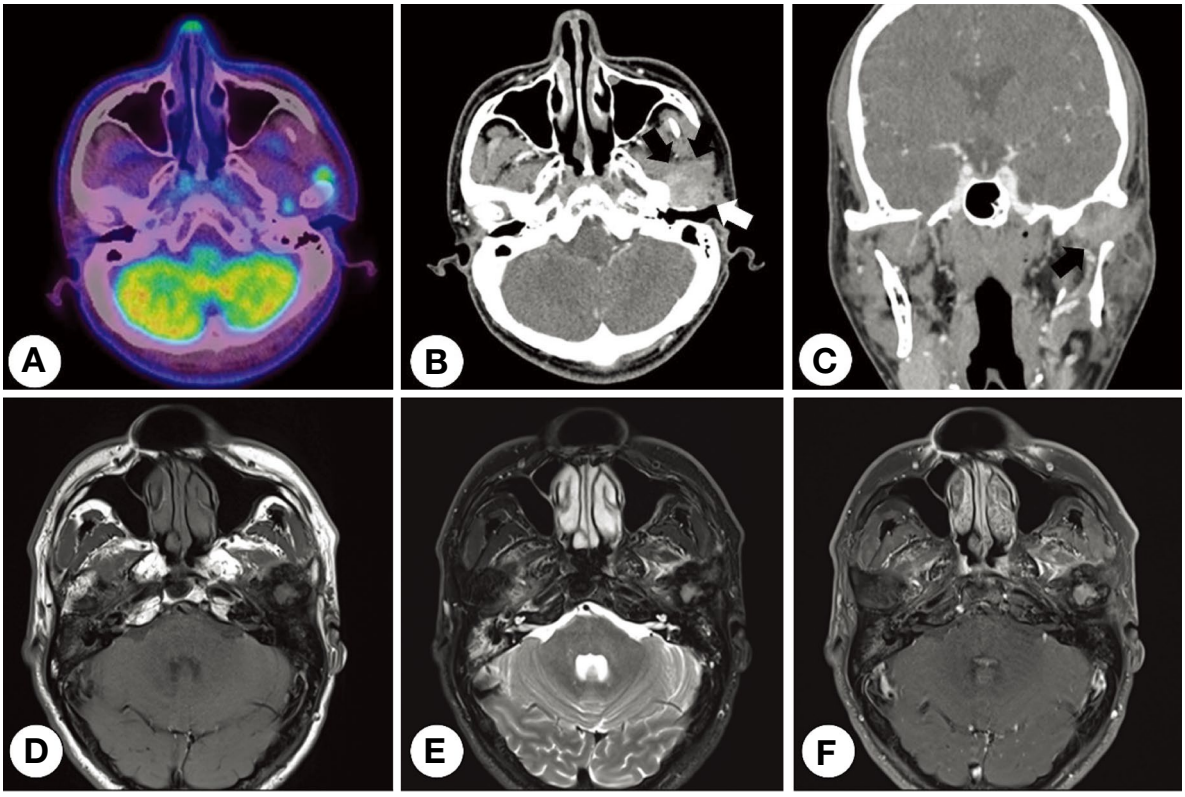


***Figure 5.*** *Imaging features of tenosynovial giant cell tumor (Case 3). A. An outside PET/CT image taken before the condylectomy shows vigorously high uptake surrounding the left mandibular condyle. B. On the contrast-enhanced axial CT image, irregular margined enhancing mass around the resected condyle was seen (black arrow). The lesion was in contact with the outer margin of the external auditory meatus (white arrow). C. The mass mentioned above with resected end of the mandibular condyle neck is shown on the coronal view of the contrast-enhanced CT image (black arrow). D-F. On Axial MR images, the lesion reveals iso signal intensity (SI) on T1 weighted, slightly high SI on T2 weighted MR images with heterogeneous enhancement. Its peripheral dark rim suggests hemosiderin deposition around tumor mass.*

10 years. The pain worsened 3-4 years before the first visit, and he complained of limited mouth opening and an open bite of the right molars. The MIO was 32 mm, with deflection to the right, and an open bite from the right first premolar to the second molar was observed.

CT imaging confirmed a soft tissue mass in the right TMJ area, and single-photon emission computed tomography and computed tomography (SPECT/CT) showed high-intensity signals in the right condyle. On the MRI image, a well-defined tumor-like mass of the anterior aspect of the right mandibular condyle was observed with abnormal fluid collection on the T2 weighted image (WI); therefore, the mass was preliminarily diagnosed as synovial chondromatosis preoperatively (Fig 3A-D).

Three months after the first visit, a mass excision was performed in the right TMJ area under general anesthesia. A preauricular approach was used, and after the incision of the lateral capsule of the TMJ, the mass in the fossa area was exposed. After removing the mass, perforation of the cranial base was not confirmed. After reduction of the anteriorly displaced disc, a Silastic™ (Dow Inc., MI, USA) interpositional silicone sheet was inserted to prevent heterotopic bone formation and postoperative adhesions. As in the previous case, self-opening exercises were taught for the patient's rehabilitation after surgery, and an occlusal stabilizing splint with pharmacological treatment was performed. The mass was histopathologically diagnosed as a TSGCT with chondroid metaplasia (Fig 4A, B).

After surgery, there were no complications like facial nerve damage or wound infection. The inserted Silastic™ was removed 5 months after the surgery. There was no tumor recurrence according to clinical and radiographic examinations, including MRI, until the last follow-up, approximately 2 years after surgery, and the last MIO was 49 mm. The patient did

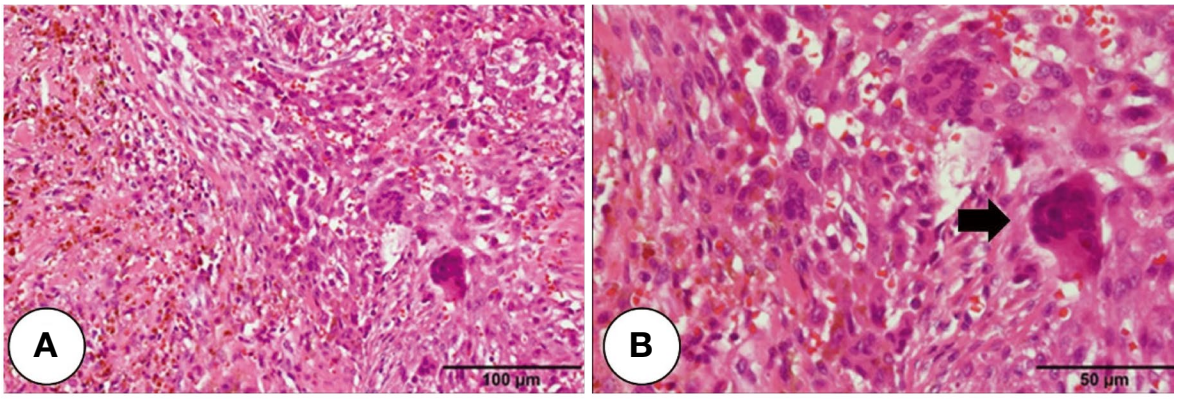


*Figure 6. Histopathologic examination (Case 3). A. B. Microphotography after Hematoxylin and eosin staining. Like in Case 2, the tumor consisted of numerous giant cells, histiocytic and spindle-shaped mononuclear cells, extravasated red blood cells, and prominent hemosiderin deposition (A: ×200, B: ×400).*

not complain of pain during mouth opening or mastication, and the movement of the mandible was symmetric.

### Case 3

A 45-year-old male patient visited our clinic for consultation with an artificial joint replacement surgery for the TMJ area. According to the patient, swelling and pain in the left preauricular area started approximately 3-4 years before, and the symptoms worsened. Three months before coming to our hospital, the patient had a confirmed tumor in the left TMJ area through imaging tests at another hospital. The position emission tomography and computed tomography (PET/CT) image showed a high signal in the left TMJ area (Fig 5A). The patient underwent mass excision and condylectomy of the left TMJ at the hospital, and the specimen was confirmed to be TSGCT.

Three months after the condylectomy, the patient did not complain of any pain or swelling in the surgical area. The patient wanted to receive treatment for masticatory discomfort and malocclusion and wanted to reconstruct his missing condyle. At our hospital, we decided to evaluate the recurrence of the tumor through CT and MRI tests approximately 6 months after the first surgery and planned TJR.

The patient underwent CT and MRI examination after 6 months. On CT, an erosive lesion was observed in the left TMJ area and infratemporal fossa. MRI showed a mass with low signal intensity on both T1 and T2 WI (Fig 5B-F). From the radiographic findings, recurrence of the TSGCT was suspected, and mass excision and simultaneous TJR of the left TMJ area under general anesthesia were decided.

A preauricular incision was made, and the recurrent mass was exposed by dissection through the scar tissue of the previous operation. After removing the mass, a retromandibular incision and dissection were performed to insert the ramal component of the Biomet® TMJ prosthesis. The ramal and fossa components were firmly fixed to the mandibular body and glenoid fossa using screws, and no complications, such as skull base perforation, were confirmed. The mass was diagnosed as a TSGCT on histopathological examination (Fig 6A, B).

Postoperatively, the patient showed favorable healing without complications, including facial nerve damage or infection. Active physiotherapy was administered for postoperative recovery, and periodic follow-up with appropriate radiographic tests was planned to confirm recurrence. Approximately 2 years have passed since the operation, and the last measured MIO was 40 mm. No specific recurrence was observed on follow-up CT and MRI images, and the patient was alive without any discomfort.

## DISCUSSION

In the three cases, surgery and rehabilitation were performed at our department due to GCLs in the TMJ area, and satisfactory results were obtained. GCLs are difficult to diagnose with a panoramic image until bony changes according to the growth of the lesion occur. Since the clinical characteristics are similar to those of common TMJ disorder, it is necessary to differentiate through additional imaging tests, including CT and MRI[9].

The patient of the second case complained pain of TMJ area with limited mouth opening, which is classic TMD symptoms. However, other pathologic conditions of the orofacial region may present symptoms mimicking TMD. For example, Marchese et al.[10] reported a pleomorphic adenoma of the parotid gland which was initially misdiagnosed as TMD because the patient complained discomfort of preauricular area. Various diseases such as Eagle's syndrome, abscess of TMJ area, benign tumors of TMJ, and malignancies including metastasis from distant organs require differential diagnosis with TMD. For differential diagnosis of atypical facial pain presenting symptoms of TMD, advanced radiological examinations such as CT for 3-dimensional observation of lesion and MRI for assessment of soft tissue will reduce misdiagnosis[11].

On CT examination, osteolysis of the affected bone around the CGCG can be observed in developed cases. The MRI characteristic of CGCG is low to intermediate signals in both T1 and T2 WI[12]. In the case of TSGCT, accumulation of hemosiderin can be seen in MRI images as a low signal area of homogenous uptake in both T1 and T2 WI with joint effusion of T2 WI[13,14]. In the second case, synovial chondromatosis was suspected before surgery due to abnormal fluid collection on MRI T2 WI. In some cases of TSGCT, differentiating it from synovial chondromatosis is challenging for similar reasons. The distinguishing point between TSGCT and synovial chondromatosis is that bone destruction due to lesion growth is observed in TSGCT[15].

According to previous studies, CGCG can be treated with surgery and/or adjuvant therapy, such as corticosteroids, interferons, or human monoclonal antibodies to receptor activator of nuclear factor kappa beta ligand (RANKL). In aggressive CGCG, surgical curettage alone may result in a high recurrence rate. Compared to curettage, surgical resection of the lesion with a 0.5 cm safety margin can reduce the recurrence rate, although damage to adjacent anatomic structures is inevitable[16]. In recurrent cases, radiation therapy and anti-angiogenesis agents can be considered[12]. Complete resection is necessary for TSGCT, but additional therapy, such as radiation therapy, may be required if the tumor is large or locally infiltrated[17]. Nicolatou-Galitis et al.[18] summarized that usage of high-dose denosumab in giant cell tumors has a role in reducing both the size of primary tumors and the pain of patients along with suppression of tumor progression. It is known that the 1-year recurrence rate is 15%, and the 5-year recurrence rate is 29% in the case of TSGCT occurring in the TMJ area[7]. Both cases reported in this study underwent complete surgical resection, and there was no evidence of recurrence until the recent follow-up visit without additional radiotherapy.

Some surgeons are reluctant to perform immediate reconstruction after tumor resection because of concerns about the artifact of MRI images to be taken for follow-up. In addition, according to the literature, patients who underwent only fat grafting of the operation area without reconstruction of the skull base or TMJ did not complain of mandibular rotation or discomfort during function[7]. However, in the guidelines for TJR in the UK in 2008, surgical resection of anatomical structures by tumor surgery was introduced as an indication for TJR[19]. There are also case reports in which a TSGCT or giant cell lesion in the TMJ area was reconstructed using a custom-made prosthesis, and satisfactory results were obtained[20,21]. In the first case, condylectomy was planned due to severe resorption of the mandibular condyle and articular fossa. Therefore, it was expected that the patient's facial asymmetry, occlusal instability, and functional discomfort would be severe after the surgery. In the last case, the affected condyle was surgically removed in a previous operation, and the patient complained about the discomfort described above. Therefore, we planned immediate reconstruction after tumor resection in both cases, and the patients lived without discomfort until 3 and 2 years after surgery, respectively. Given this, immediate reconstruction with alloplastic TJR can be recommended if the condyle is to be resected during surgical removal of the GCL in the TMJ area to reduce postoperative discomfort and improve the quality of life of patients.

If proper physical therapy is not done after open surgery of the TMJ, there may be atrophy of the masticatory muscle due to non-use after surgery, and there is a risk of ankylosis and adhesions of the TMJ due to prolonged immobilization[22]. Also, clonazepam, diazepam, and muscle relaxants are recommended because clenching or bruxism may worsen depending on the change in occlusion after surgery[23]. In our department, we educate all patients undergoing TMJ open surgery on implementing active mouth opening after surgery. In some patient groups, we teach to force the opening by inserting own fingers. In addition, moist hot packs are recommended for the chin, neck, and shoulder areas. Central muscle relaxants (afloqualone), NSAIDs, and diazepam are prescribed if the discomfort is severe. All patients presented in this case report were treated with the physiotherapy and pharmacological treatments described above to reduce the

possibility of postoperative complications and to control pain.

In conclusion, although GCL is rare in the TMJ, it can invade the cranial base by lesion growth and result in severe functional damage by resorbing the mandibular condyle and temporal bone. After surgical removal, depending on the resection range, immediate reconstruction with TJR can be performed to reduce postoperative discomfort and improve the patient's quality of life. Appropriate physiotherapy and medications are required after surgery to reduce complications. Finally, a periodic follow-up to check for recurrence is recommended with appropriate imaging tests.

## DISCLOSURE STATEMENTS

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

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