Computed tomography and magnetic resonance imaging characteristics of giant cell tumors in the temporomandibular joint complex

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ABSTRACT

Purpose: This study aimed to investigate the computed tomography and magnetic resonance imaging features of giant cell tumors in the temporomandibular joint region to facilitate accurate diagnoses.

Materials and Methods: From October 2007 to June 2020, 6 patients (2 men and 4 women) at Yonsei University Dental Hospital had histopathologically proven giant cell tumors in the temporomandibular joint. Their computed tomography and magnetic resonance imaging findings were reviewed retrospectively, and the cases were classified into 3 types based on the tumor center and growth pattern observed on the radiologic findings.

Results: The age of the 6 patients ranged from 25 to 53 years. Trismus was found in 5 of the 6 cases. One case recurred. The mean size of the tumors, defined based on their greatest diameter, was 32 mm (range, 15-41 mm). The characteristic features of all cases were a heterogeneously-enhancing tumorous mass with a lobulated margin on computed tomographic images and internal multiplicity of signal intensity on T2-weighted magnetic resonance images. According to the site of origin, 3 tumors were bone-centered, 2 were soft tissue-centered, and 1 was peri-articular.

Conclusion: Computed tomography and magnetic resonance imaging yielded a tripartite classification of giant cell tumors of the temporomandibular joint according to their location on imaging. This study could help clinicians in the differential diagnosis of giant cell tumors and assist in proper treatment planning for tumorous diseases of the temporomandibular joint. (Imaging Sci Dent 2021; 51: 149-54)

KEY WORDS: Diagnostic Imaging; Giant Cell Tumors; Magnetic Resonance Imaging; Temporomandibular Joint

Introduction

Giant cell tumor (GCT) is regarded as benign intraosseous or extraosseous neoplasms arising from multinucleated giant cells. ¹⁻³ It can be divided into 3 entities depending on their origin: GCT of the bone, GCT of the soft tissue, and tenosynovial GCT (TGCT), which encompasses pigmented villonodular synovitis and GCT of the tendon sheath according to the 2013 World Health Organization classification.³

The histologic appearance of GCT varies depending on the proportion of synoviocytes, histiocytes, osteoclast-like giant cells, and xanthomatous histiocytes, the amount of

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hemosiderin, and the degree of collagenization.^{1,4,5} The high degree of variation in cellular composition makes it difficult to diagnose GCT accurately based on its histologic appearance, especially in biopsy specimens.⁴⁻⁷

The temporomandibular joint (TMJ) plays a pivotal role in articulation between the mandible and middle cranial fossa. The structure of the TMJ is complex, including the disc, retrodiscal tissue, synovium, and synovial fluid; furthermore, it is surrounded by masticatory muscles, tendons, and ligaments to achieve efficient translation. When a GCT involves the TMJ region, this anatomical complexity makes the diagnosis more challenging than for GCT in large joints such as the hip and shoulder. This difficulty is augmented by the fact that GCT in the TMJ region is very infrequent. Even including cases reported using other synonyms, lesser than 100 GCTs in the TMJ have been reported in the English-language literature. In particular, reports of GCT in the

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mandibular condyle are extremely rare. 14,19

The purpose of this study was to investigate the radiologic features of GCTs of the TMJ on computed tomography (CT) and magnetic resonance imaging (MRI) to facilitate accurate diagnoses.

Materials and Methods

A retrospective review, approved by Institutional Review Board of Yonsei University Dental Hospital, was done to evaluate the clinical and radiologic features of 6 patients with histologically proven GCT in the TMJ. The patients were recruited through an electronic search of our institution's pathological database from October 2007 to June 2020. The clinical records of the enrolled patients were thoroughly reviewed by a radiologist with over 10 years of experience in oral and maxillofacial radiology. The CT scans of the 6 patients were obtained by multi-detector CT (Optima CT 520; Philips Medical Systems, Cleveland, OH, USA). They also underwent MRI scans using a 3.0-T unit (Pioneer; GE Healthcare, Waukesha, WI, USA). The MRI scans included multiplanar spin-echo T1-weighted imaging (TR/TE, 734/9) and multiplanar spin-echo T2-weighted imaging (TR/TE, 2707/66).

For each case, CT and MRI findings were evaluated with an emphasis on the lesion's size, margin, effect on the adjacent tissue, internal attenuation on CT, and signal intensity on MRI. The size of the lesion was calculated based on mean maximum diameter measured in 3 orthogonal planes. The margin of the lesion was classified as round to ovoid, lobulating, or infiltrative. Two categories were used to describe bone structure changes (remodeling or destruction), the former of which included cortical thinning, expansion, or bone erosion. The signal intensity of the lesion on T1- and T2-weighted MRI was compared with that of the masticator muscle by visual assessment. On post-contrast CT and MRI, the pattern of enhancement in the solid portions of the lesion was categorized as homogeneous or heterogeneous.

Based on these assessments, all cases were categorized as bone-centered, soft tissue-centered, or peri-articular types, using a modified version of a classification system proposed in a previous study.²⁴ The bone-centered type was defined as a lesion confined to the condyle or temporal bone. The soft tissue-centered type seemed to originate from the soft tissue around the TMJ, and presented no destruction of the neighboring bony structure. In the peri-articular type, the lesion originated from the connective tissue outside of the mandibular condyle with invasion into the bone marrow space (Fig. 1).

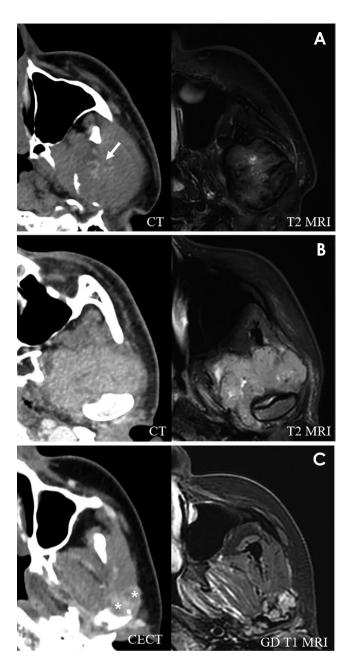


Fig. 1. Giant cell tumors in the temporomandibular joint region are categorized as bone-centered (A), soft tissue-centered (B), or peri-articular (C) according to the tumor's center and growth pattern. The right and left figures indicate a tumor mass on magnetic resonance imaging and computed tomography. Bone-centered type (A) shows a lobulated tumorous mass with internal high attenuation portion (white arrow). Peri-articular type (C) presents bony destruction by infiltrative tumorous mass (white asterisk).

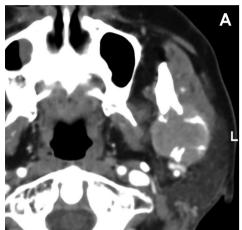
Results

The patients' demographic characteristics, initial symptoms, and clinical findings are summarized in Table 1. The patients consisted of 2 men and 4 women, with an age range of 25-53 years (mean age, 32.8 years). The left TMJ

Table 1. Summary of the clinical and demographic characteristics of the 6 patients

No. of	C	Age	C: 1-		Clinical sig	ns		Follow-up	
case	Sex	(years)	Side	Pain	Swelling	Trismus	Treatment	Recurrence	Duration (months)
1	Male	29	Left	Yes	Yes	Yes	Surgical excision	_	1
2	Female	53	Left	Yes	Yes	No	Surgical excision	No	121
3	Male	28	Left	Yes	No	Yes	Surgical excision	No	49
4	Female	30	Right	Yes	Yes	Yes	Surgical excision	No	39
5	Female	25	Right	No	No	Yes	Surgical excision	Yes	91
6	Female	32	Left	No	No	Yes	Surgical excision	No	6

No.: number



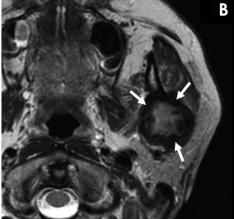


Fig. 2. A. A bone-centered giant cell tumor is observed as a lobulated hyperdense tumorous mass on the left condylar head. B. At the periphery, a low signal intensity on T2-weighted magnetic resonance imaging is detected (white arrow).

was affected in 4 cases and the right TMJ in the other 2 cases. The patients complained of trismus (83.3%), pain (66.7%), and facial swelling (50.0%). All patients underwent complete surgical excision without postoperative radiation or chemotherapy and were followed up for at least 1 month, with the longest follow-up being 121 months (mean follow-up duration, 51 months). One patient underwent additional resection surgery for removal of a residual or recurrent lesion after 1 year. No recurrence was reported in the other 5 patients during the postoperative follow-up period.

The CT and MRI features of the lesions are summarized in Table 2. The mean size of the tumors, defined based on their greatest diameter, was 32 mm (range, 15-41 mm). Three tumors were bone-centered, 2 were soft tissue-centered, and 1 was peri-articular.

The CT images of the bone-centered tumors presented cortical expansion of the mandibular condyle and lobulated septation, suggesting that the lesions had a lobulated margin. Two of the 3 cases showed low signal intensity at the periphery on both T1 and T2-weighted MRI sequences. The central portion of the lesions, unlike the peripheral portion, showed a variety of signal intensity (Fig. 2). Of partic-

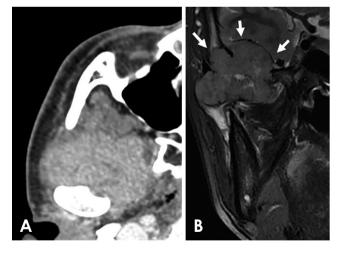


Fig. 3. A. The soft tissue-centered type of giant cell tumor, shown as a heterogeneously enhancing tumorous mass, is located in the masticatory space on computed tomography. B. The tumor is observed as iso or slightly high signal intensity compared to the masticatory muscle on coronal T1-weighted magnetic resonance imaging with intracranial extension (white arrow).

ular note, the T2-weighted MRI scans showed high T2 signal intensity with internal chondroid metaplasia (case 1). In



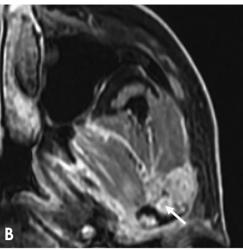


Fig. 4. A. The peri-articular type of giant cell tumor presents bony remodeling of the left condyle's anterior border on a panoramic radiograph (white triangle). B. On axial contrast-enhanced T1-weighted magnetic resonance imaging, the tumorous mass shows an infiltrative growth pattern into the condylar head with heterogeneous enhancement (white arrow).

contrast, the soft tissue-centered type showed a well-encapsulated tumorous mass with heterogeneous enhancement on contrast-enhanced CT and iso or slightly high signal intensity compared to the masticatory muscle on T1-weighted MRI sequences (Fig. 3). Its growth pattern was depicted as a widened joint space and/or compression of the neighboring masticatory muscles with minimal potential for bony destruction. The peri-articular type had an infiltrative growth pattern that was presumed to have multiple centers along the synovium or other connective tissue (Fig. 4).

Discussion

GCT is a locally aggressive neoplasm originating from undifferentiated mesenchymal cells.¹⁻³ Due to its neoplastic etiology, which may be still controversial, it is pathologically distinguishable from giant cell reparative lesion (GCRL).⁶ It can be classified according to whether it originates in the bone marrow, soft tissue (e.g., muscle), or the synovial lining in the joint space.³ However, due to the complex anatomic structures of the TMJ, it is often difficult to accurately categorize the tumor, especially if the lesion is very large and aggressive.^{6,7,16}

GCT of the bone usually originates from the medullary space of long bones, and in the head and neck region it predominantly affects the sphenoid, ethmoid, and temporal bones. GCT of the bone has a male predilection, ^{2,3,8,14,19} whereas TGCT, which usually arises from the joint synovium, presents no definite sex predilection. ^{1,4,11,15,20,22} However, the present study of TMJ GCT showed a female predilection, with a male-to-female ratio of 1:2.

Although GCT can be found in any age group, it most commonly occurs in the third or fourth decade of life, which is consistent with our findings.^{19,22} This is unlike GCRL, which is found in both jaws with a roughly even distribution and tends to occur in the second decade of life.^{5,7}

The most common presenting symptoms of GCT include a pre-auricular palpable mass or swelling, pain, trismus, or hearing loss. 9-25 The symptoms are non-specific to the lesion, and vary depending on the anatomical structures that the tumor involved. However, in the present study, the chief complaint differed according to the subtype of GCT. Bone-centered tumors tended to be accompanied by pain, while patients with soft tissue-centered tumors reported trismus. This might be attributed to the growth potential of the tumor mass extending into the masticatory muscle.

The 3 subtypes of GCT in the TMJ region showed different CT and MRI features. The bone-centered type occurred in 3 cases (cases 1-3), of which 1 was centered in the temporal bone and the others were located in the condylar head. In one of the latter cases, intracranial extension was detected. The imaging features of bone-centered osteolytic GCT were a lobulated margin with cortical thinning and expansion of the condylar head, and intracranial extension. On T2-weighted MRI, its signal intensity was observed as internal patch-like multiplicity with a peripheral dark rim.

The soft tissue-centered type (cases 4 and 5) showed a heterogeneously enhancing mass accompanied by compressive bony remodeling, sclerosis, and periosteal new bone formation. These tumors presented heterogeneous signal intensity on T2-weighted images and slightly high signal intensity on T1-weighted images. They showed a mass effect, including joint space widening and intracranial extension, depending on the tumor size and center. These imaging findings are similar to those of leiomyosarcoma or fibromyxosarcoma, meaning that a precise differential diagnosis is required.

cable 2. Computed tomographic (CT) and magnetic resonance imaging (MRI) features of giant cell tumors with involvement of the temporomandibular joint region

No. of	Mean size	E		Effect	Effect on the adjacent structure	cture	Internal enhancement of CT	rnal ent of CT	MRI signal intensity	1 intensity
case	(mm)	lype	Margin	Bony change	Widening of the joint spaces	Intracranial extension	Degree	Pattern	T1-weighted	T2-weighted
1	41	Bone-centered	Lobulated	Remodeling	No	No	Minimal	Hetero	Low at periphery	Low at periphery
2	35	Bone-centered	Lobulated	Remodeling	Yes	Yes	High	Hetero	Low at periphery	Low at periphery
3	32	Bone-centered	Lobulated	Remodeling	Yes	Yes	High	Hetero	Iso	High
4	39	Soft tissue-centered	Lobulated	Remodeling	Yes	Yes	High	Hetero	Moderate	Moderate to high
5	30	Soft tissue-centered	Lobulated	Remodeling	No	No	High	Hetero	High	Moderate
9	15	Peri-articular	Infiltrative	Destruction	No	No	High	Hetero	Iso	Low to high

The peri-articular type (case 6) showed an infiltrative growth pattern into the bone marrow from the tendon sheath or synovium outside the bony part of the TMJ complex. Although there may be some controversy in the classification, this type can be regarded as diffuse pigmented villonodular synovitis, which has rarely been reported in the TMJ region.

Kim et al. proposed that GCT shows a dark signal intensity on T1 and T2 and the blooming effect, suggesting internal hemosiderin deposition. ^{12,22,24} However, these characteristics were not clearly detected in the present study.

The differential diagnosis of GCT includes aneurysmal bone cyst, chondroblastoma, chondrosarcoma, giant cell reparative granuloma, and the aggressive type of synovial chondromatosis. 5-7,24,26 In cases of internal calcification, it is necessary to distinguish GCT from bone-forming malignancies. 18,24,27,28 Furthermore, when an aggressive growth pattern is evident, the possibility of malignancy cannot be ruled out until the diagnosis is confirmed through a histologic examination. 7,18,24 The imaging features of GCT shown by CT and MRI can facilitate an accurate diagnosis and suggest optimal treatment.

In conclusion, the CT and MRI characteristics of GCT in the TMJ varied depending on the location in the present study. This information may be helpful for diagnosing GCT in the TMJ area.

Conflict of Interest: None

References

- 1. Goldblum JR, Folpe AL, Weiss SW. Enzinger & Weiss's soft tissue tumors. 7th ed. Philadelphia: Elsevier; 2020.
- Jo VY, Hornick JL. Neoplastic mimics in soft tissue and bone pathology. New York: Demos Medical Pub.; 2016.
- Fletcher CD, Bridge JA, Hogendoorn PC, Martens F. World Health Organization classification of tumours of soft tissue and bone. 4th ed. Lyon: IARC Press; 2013.
- Lindberg MR, Lucas D, Cassarino D, Gardner JM, Stallings-Archer K. Diagnostic pathology. Soft tissue tumors. 3rd ed. Philadelphia: Elsevier; 2019.
- Lee JC, Huang HY. Soft tissue special issue: giant cell-rich lesions of the head and neck region. Head Neck Pathol 2020; 14: 97-108.
- Silvers AR, Som PM, Brandwein M, Chong JL, Shah D. The role of imaging in the diagnosis of giant cell tumor of the skull base. AJNR Am J Neuroradiol 1996; 17: 1392-5.
- Hamza A, Gidley PW, Learned KO, Hanna EY, Bell D. Uncommon tumors of temporomandibular joint: an institutional experience and review. Head Neck 2020; 42: 1859-73.
- 8. Som PM, Curtin HD. Head and neck imaging. 5th ed. St. Louis: Mosby; 2011. p. 1588-9.

- Lapayowker MS, Miller WT, Levy WM, Harwick RD. Pigmented villonodular synovitis of the temporomandibular joint. Radiology 1973; 108: 313-6.
- 10. Bemporad JA, Chaloupka JC, Putman CM, Roth TC, Tarro J, Mitra S, et al. Pigmented villonodular synovitis of the temporomandibular joint: diagnostic imaging and endovascular therapeutic embolization of a rare head and neck tumor. AJNR Am J Neuroradiol 1999; 20: 159-62.
- 11. Lee JH, Kim YY, Seo BM, Baek SH, Choi JY, Choung PH, et al. Extra-articular pigmented villonodular synovitis of the temporomandibular joint: case report and review of the literature. Int J Oral Maxillofac Surg 2000; 29: 408-15.
- 12. Kim KW, Han MH, Park SW, Kim SH, Lee HJ, Jae HJ, et al. Pigmented villonodular synovitis of the temporomandibular ioint: MR findings in four cases. Eur J Radiol 2004: 49: 229-34.
- Herman CR, Swift JQ, Schiffman EL. Pigmented villonodular synovitis of the temporomandibular joint with intracranial extension: a case and literature review. Int J Oral Maxillofac Surg 2009; 38: 795-801.
- 14. Roberts DS, Faquin WC, Deschler DG. Giant cell tumors of the temporal bone and infratemporal fossa: a case report and review of the literature. Laryngoscope 2010; 120 Suppl 4: S180.
- 15. Yoon HJ, Cho YA, Lee JI, Hong SP, Hong SD. Malignant pigmented villonodular synovitis of the temporomandibular joint with lung metastasis: a case report and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011; 111: e30-6.
- Liu YK, Chan JY, Chang CJ, Huang JS. Pigmented villonodular synovitis of the temporomandibular joint presenting as a middle cranial fossa tumor. J Oral Maxillofac Surg 2012; 70: 367-72.
- Le WJ, Li MH, Yu Q, Shi HM. Pigmented villonodular synovitis of the temporomandibular joint: CT imaging findings. Clin Imaging 2014; 38: 6-10.
- 18. Pina S, Fernandez M, Maya S, Garcia RA, Noor A, Pawha PS, et al. Recurrent temporal bone tenosynovial giant cell tumor with chondroid metaplasia: the use of imaging to assess recurrence. Neuroradiol J 2014; 27: 97-101.

- Byun JH, Park KB, Ko JS, Ahn SK. Giant cell tumor of infratemporal fossa and mandibular condyle: a case report. J Int Adv Otol 2015; 11: 88-91.
- 20. Safaee M, Oh T, Sun MZ, Parsa AT, McDermott MW, El-Sayed IH, et al. Pigmented villonodular synovitis of the temporomandibular joint with intracranial extension: a case series and systematic review. Head Neck 2015; 37: 1213-24.
- Chen Y, Cai XY, Yang C, Chen MJ, Qiu YT, Zhuo Z. Pigmented villonodular synovitis of the temporomandibular joint with intracranial extension. J Craniofac Surg 2015; 26: e115-8.
- Carlson ML, Osetinsky LM, Alon EE, Inwards CY, Lane JI, Moore EJ. Tenosynovial giant cell tumors of the temporomandibular joint and lateral skull base: review of 11 cases. Laryngoscope 2017; 127: 2340-6.
- 23. Brant JA, Kaufman AC, Luu N, Grady SM, O Apos Malley BW, Ruckenstein MJ. Pigmented villonodular synovitis presenting as unilateral hearing loss: review of the literature and case report. ORL J Otorhinolaryngol Relat Spec 2019; 81: 171-82.
- 24. Wang JG, Liu J, He B, Gao L, Zhang L, Liu J. Diffuse tenosynovial giant cell tumor around the temporomandibular joint: an entity with special radiologic and pathologic features. J Oral Maxillofac Surg 2019; 77: 1022.e1-39.
- 25. Liu Y, Fan BH, Tan YR, Zhu DW, Dong MJ, Wang LZ, et al. Diffuse-type tenosynovial giant cell tumor of the temporomandibular joint with skull base invasion: a report of 22 cases with literature review. Oral Surg Oral Med Oral Pathol Oral Radiol 2021; 131: 16-26.
- 26. Gigliotti J, Alghamdi O, El-Hakim M, Makhoul N. Central giant cell granuloma of the mandibular condyle: a case report, literature review, and discussion of treatment. Oral Maxillofac Surg Cases 2015; 1: 42-6.
- Oh KY, Yoon HJ, Lee JI, Hong SP, Hong SD. Chondrosarcoma of the temporomandibular joint: a case report and review of the literature. Cranio 2016; 34: 270-8.
- 28. Anbinder AL, Geraldo BM, Guimarães Filho R, Pereira DL, Almeida OP, Carvalho YR. Chondroid tenosynovial giant cell tumor of the temporomandibular joint: a rare case report. Braz Dent J 2017; 28: 647-52.