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Synovial chondromatosis of the temporomandibular joint with 400 loose bodies: a case report and literature review

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Abstract

Synovial chondromatosis (SC) is a benign condition characterized by the formation of metaplastic cartilage in the synovial membrane of the joint, resulting in numerous attached and unattached osteocartilaginous bodies. SC mostly affects the large synovial joints, especially the knee, hip, elbow, and ankle, whereas involvement of the temporomandibular joint (TMJ) is rare. Approximately 240 cases of SC of the TMJ have been reported in the English-language literature to date. The number of loose bodies varies among patients but usually ranges from the dozens to around 100. We herein report a case of SC of the TMJ accompanied by approximately 400 loose bodies in a healthy 53-year-old woman. Such a high number of loose bodies within a small space is extremely rare. We also include a brief discussion about the differential diagnoses and current diagnostic approaches to SC of the TMJ. Notably, delayed diagnosis or misdiagnosis is common because of the nonspecific nature of the presenting complaints.

Keywords

Synovial chondromatosis, temporomandibular joint, loose body, differential diagnosis, case report, literature review

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Introduction

Synovial chondromatosis (SC) rarely occurs in the temporomandibular joint (TMJ). Approximately 240 such cases have been reported in the English-language literature

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to date,[1] but the etiology has not been completely clarified. The usual clinical signs and symptoms are restricted movement of the mandible, pain, preauricular swelling, and crepitation. Delayed diagnosis or misdiagnosis is common because of the nonspecific nature of the presenting complaints and the low incidence rate.[2] In this report, we present an uncommon case of SC of the TMJ accompanied by approximately 400 loose bodies. We also discuss the differential diagnoses and current diagnostic approaches to SC of the TMJ.

Case presentation

A 53-year-old woman presented to our department in June 2011 with a left preauricular swelling. Her chief complaint was that the swelling had been slowly growing for about 5 months. Additionally, she complained of pain in the left TMJ area; the ability to open her mouth was limited, and each time she opened her mouth widely, she felt a clicking sensation in the left preauricular area. The patient had no history of trauma, rheumatoid arthrosis, or septic arthritis. She revealed that her left TMJ had become dislocated about 2 years previously, and the patient had repositioned it herself; however, there had been no subsequent recurrence. Before she presented to our department, she visited a local hospital for treatment and underwent a computed tomography (CT) scan of the TMJ region (Figure 1). Because this was the first time the clinicians at that hospital had encountered such a case, the patient was referred to our hospital. Physical examination revealed a preauricular immovable and tender mass measuring approximately 20×30 mm, with medium to somewhat soft firmness. Her maximal mouth opening was limited to 25 mm, with slight deviation to the left side. No facial nerve paralysis or hearing disturbance was detected.

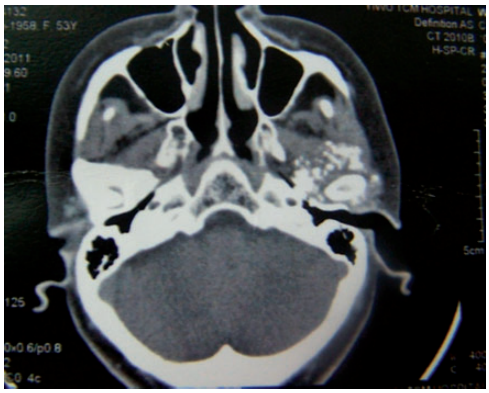

Figure 1. Preoperative axial computed tomography scan of the temporomandibular joint region. The image shows radiodense bodies surrounding the left condyle.

A CT scan showed multiple small radiodense particles surrounding the left condyle, especially in the anterior region of the left TMJ capsule. Magnetic resonance imaging (MRI) was performed in our hospital to check the condition of the soft tissues surrounding the lesion. This examination showed an expanded high-signal area, including the anterior and lateral space of the left condyle, containing multiple signal-void particles (Figure 2). The TMJ disc was in the clinically normal position and no perforation was suspected, and the surface of the left condyle was smooth. According to the physical examination and radiological findings, the preoperative diagnosis was SC of the left TMJ. The patient consented to undergo hospitalization and surgical treatment. After being admitted to the inpatient department, the patient underwent arthrotomy under general anesthesia; the preauricular approach was chosen to gain access to the left TMJ. When the TMJ capsule was opened, it discharged a viscous fluid containing many similarly sized opalescent, glistening loose bodies (Figure 3(a)). Most of the loose bodies were collected, although some were initially sucked into the suction unit. The disc was in the correct

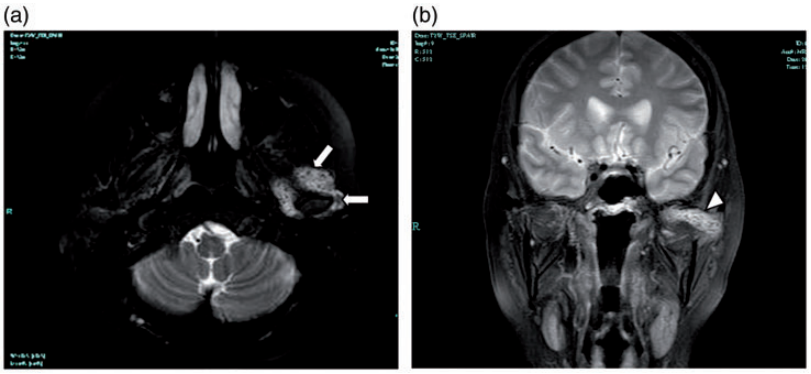

Figure 2. Magnetic resonance imaging (MRI) of the temporomandibular joint region showing the obviously expanded joint space. (a) Axial MRI revealing expansion of the anterior joint space (white arrow). (b) Coronal MRI showing the upper boundary of the mass (white arrowhead).

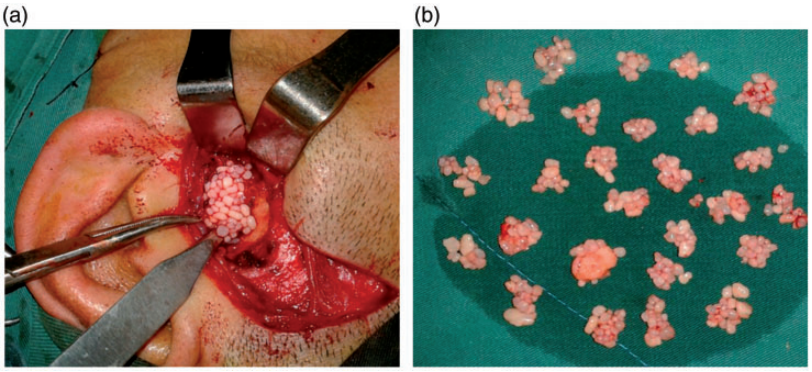

Figure 3. Surgical views of the left temporomandibular joint (TMJ). (a) During the arthrotomy procedure, numerous loose bodies could be seen after the TMJ capsule was opened. (b) Loose bodies removed from the left TMJ capsule (not including the bodies that were lost to the suction unit).

position without perforation, and the condylar head was smooth; therefore, only a partial synovectomy procedure was performed at the affected region of the synovium. The cavity was irrigated with a large volume of saline solution. After surgery, we counted up to 400 calcified bodies (Figure 3(b)), with the largest measuring approximately 8×5 mm.

Histological examination revealed that the excised synovial membrane was chondrometaplastic and that the calcified loose bodies were composed of hyaline cartilage covered by fibrous connective tissue (Figure 4). The histological findings confirmed the diagnosis of SC of the left TMJ. A postoperative CT scan showed no remaining loose bodies (Figure 5). The patient recovered well after surgery, and no facial paralysis or hearing loss was noted. At the 3-month follow-up appointment, physical examination revealed no mandibular deviation, malocclusion, or preauricular tenderness, and her maximal incisal opening was 35 mm. We conducted follow-up every year after

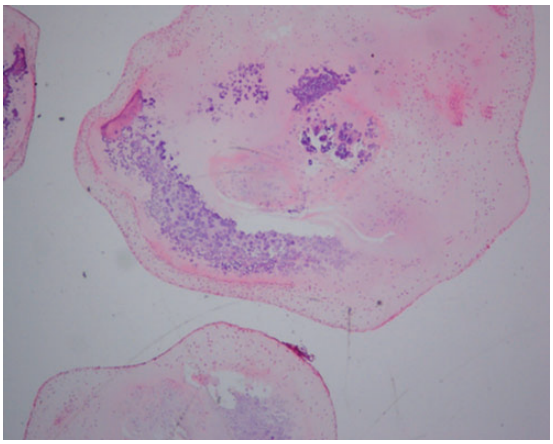

Figure 4. Histological examination of the loose bodies. Fibrous connective tissue can be seen (hematoxylin–eosin, original magnification ×100).

Figure 5. Postoperative axial computed tomography scan of the temporomandibular joint region. The image shows the normal structure of the left condyle: there were no remaining particles.

surgery for 9 years and found no signs of recurrence.

Discussion

SC is an uncommon benign lesion of the synovial joint that is especially rare in the TMJ, where it is seen in only 3% of reported cases.[3] There is a clear female predominance for the condition.[4] Its etiology remains elusive, but inflammation, trauma, and joint overuse have been cited as potential causative factors. The predominant clinical signs and symptoms are restricted movement of the mandible, pain, preauricular swelling, and crepitation.[5] To establish a primary diagnosis of SC of the TMJ, the patient's complaint history, clinical signs, and symptoms should be considered along with the radiographic findings. This condition is easily misdiagnosed using conventional X-ray examinations because the small cartilage bodies are usually uncalcified or are <1 mm in diameter.[6] When bone destruction is present at the top of the joint fossa, CT examination is particularly important because it helps to determine the scope and extent of bone destruction at the bottom of the middle cranial fossa and whether the lesion has violated the brain. For visualization of loose bodies with a good degree of calcification, CT examination is far superior to conventional X-ray examination. High-resolution CT can even detect calcified loose bodies of <1 mm. However, for loose bodies with a poor degree of calcification, CT is unsatisfactory or ineffective.[7] Cone beam CT, which has a smaller radiation dose than spiral CT, can also clearly show bone

changes in the joint area and display calcified loose bodies; however, it is less effective than spiral CT at displaying soft tissues.[8] MRI can show changes in the structure of the TMJ, including the position of the joint capsule, joint discs, and joint cavity effusion; it can also show loose bodies with a low degree of calcification and invasion of adjacent structures by SC. The characteristic appearances on proton-weighted images and T2 images have important reference value for the diagnosis of SC,[9] which mainly manifests as joint cyst enlargement, joint cavity effusion, and synovial thickening. The typical "circular" image can be seen in the lesions, with cartilage nodules or calcified masses at the center showing low signal intensity and the surrounding tissue showing high signal intensity due to fibrous tissue or effusion. Liu et al.[10] evaluated the diagnostic accuracy of MRI for SC of the TMJ and found that the incidence of MRI-diagnosed SC was in accordance with that of arthroscopic and open surgery. They recommended MRI as a relatively noninvasive and effective diagnostic modality for detecting SC. Additionally, most authors report CT and MRI as the most useful imaging techniques for diagnosis and surgical planning, especially considering that SC can spread into the cranial space.[11] Fine needle aspiration biopsy (FNAB) is a valuable tool in the diagnosis of head and neck lesions,[12] but not all cases of SC can be accurately diagnosed by FNAB because of inadequate tissue. Ardekian et al.[5] reported that FNAB was diagnostic for SC in four of nine cases. In their opinion, this result was not surprising because samples of cartilage and bone are difficult to obtain with a small-gauge needle, and FNAB has limited diagnostic value for the evaluation of fluid-filled and cystic spaces. Arthroscopy is another effective procedure for the diagnosis of TMJ diseases.[13] Nevertheless, because of the invasive nature of the procedure, it may not be the first choice as a diagnostic tool. All of these diagnostic techniques have advantages for the diagnosis of SC of the TMJ, but a definitive diagnosis can only be made on the basis of a histological examination of the loose bodies or synovial membrane.[14]

In 1977, Milgram[15] described three stages of the progression of SC (Table 1). The main pathological feature of SC of the TMJ is the presence of cartilage nodules adjacent to the synovium, usually connected only by the stalk.[16] The central area of the loose bodies loses its nutrient source, undergoes necrosis, and calcifies. Yoshida et al.[17] revealed the morphological properties of loose bodies of the TMJ by scanning electron microscopy. In the inside portion, the collagen fibers ran very densely in the same direction in an orderly manner. However, the outside portion seemed to exhibit a porous pattern. This might be explained by the surface and outside portion containing many active fibroblasts. As a result, it seems that loose bodies might develop in a multi-layer manner in which fibrous tissues become loosely piled up around the inside portion. The most commonly used antigens in immunohistochemistry are proliferating cell nuclear antigen and Ki-67.[18] Both are important for detecting the disease activity at the time of diagnosis. Proliferating cell nuclear antigen is usually high in the proliferative stage (Milgram stages 1 and 2), and Ki-67 is usually low. Li et al.[19] reported that in patients with SC of the TMJ, transforming growth factor β3 (TGF-β3) is expressed in the synovium of the TMJ and loose bodies. TGF-β3 upregulates the expression of cartilage, osteogenesis, and angiogenesis genes in SC synovial cells. Fibroblast growth factor 2 upregulates the expression of TGF-β3. Therefore, TGF-β3 is a pathogenetic factor of TMJ SC. These two cytokines not only increase the TMJ synoviocytes' expression of the Sox9 and Wnt4 genes,

Table 1. Milgram histological classification.

Stage	
1	A lesion without detached bodies
2	A lesion with synovial metaplasia and presence of loose bodies
3	A lesion with loose bodies and intact synovium

which are potent regulators of the chondrocyte phenotype and regulate the gene expression of aggrecan, but also increase the expression of vascular endothelial growth factor A, which is a potent angiogenic factor.[20] Yoshida et al.[21] reported that Ki-67 expression was not detected in almost all cases of loose bodies but that mild expression of Ki-67 was detected on the synovium. These results indicate that although the loose bodies released into the joint cavity do not have independent proliferative activity, the synovium may play a very important role in the proliferation of the loose bodies in patients with SC.

Many authors have noted that misdiagnoses are common because of the low incidence of SC of the TMJ and the nonspecific nature of the presenting complaints.[22,23] The similarities and differences between SC of the TMJ and other lesions should be thoroughly understood to achieve an accurate primary diagnosis of this disorder. SC of the TMJ accompanied by preauricular swelling should be distinguished from a parotid mass. In terms of clinical symptoms, the former usually leads to TMJ disorders such as limitation of jaw movement, sound in the TMJ, and chewing discomfort. In contrast, a parotid mass is not usually clinically uncomfortable unless it is malignant.

The existence of loose bodies in the TMJ is the most characteristic radiographic feature of SC of the TMJ, but other signs may also occur. Lesions can also result in intracapsular condylar fractures, degenerative joint diseases, tuberculous arthritis, and other complications.[24,25] Therefore, it is difficult to make an accurate diagnosis by relying exclusively on radiological findings. In the present report, we have organized these lesions into four categories (Table 2), which may be helpful to understand the differential diagnoses for SC of the TMJ. The main differential diagnoses include osteoarthritis, calcium pyrophosphate deposition disease (CPDD) or pseudogout, osteochondroma, pigmented villonodular synovitis, and osteochondritis. Osteoarthritis is an age-related degenerative disease that is common in women; on imaging, it often manifests as osteophyte formation, bone destruction, flattening of the articular nodules, bone sclerosis of the joint socket, and joint space narrowing.[8] Compared with SC, the calcified corpuscles that form in osteoarthritis are smaller, and the mandibular condyle and glenoid bone can appear normal in patients with SC. CPDD, or pseudogout, is arthritis characterized by the precipitation of calcium pyrophosphate crystals. This condition is common in elderly patients but rarely involves the TMJ. The main X-ray manifestation of CPDD is cartilage calcification, which is similar to the pathological and imaging manifestations of SC; however, CPPD of the TMJ is rare and usually has related predisposing factors (gout).[26] Osteochondroma is common in the limb bones and rarely affects the TMJ area, and CT examination often exhibits continuity between the original bone and the cortical contour of the tumor, which is not observed in patients with SC.[27] Pigmented villonodular synovitis and osteochondritis are reactive focal calcifications and should be included as

Table 2. Differential diagnoses for of synovial chondromatosis of the TMJ.

Tumor and Tumor-like lesions	Cysts	Inflammatory lesions	Others
Osteochondroma	Simple bone cyst	Pigmented villonodular synovitis	Langerhans cell histiocytosis
Osteoma	Epidermal inclusion cyst	Tuberculous arthritis	Internal derangement of TMJ
Osteoblastoma	Dermoid and epidermoid cyst	Osteochondritis dissecans	Degenerative joint disease
Osteosarcoma	Sebaceous cyst	Parotitis	Chondrocalcinosis
Plasma cell myeloma	Synovial cyst		Benign hypertrophy of condyle
Non-ossifying fibroma	Parotid cyst		
Tumors related to parotid gland	Ganglion		
	Aneurismal bone cyst		

TMJ, temporomandibular joint.

differential diagnoses.[28,29] Large numbers of calcified bodies will not appear in these diseases.

Conclusion

We have reported an uncommon case of SC of the TMJ accompanied by approximately 400 loose bodies in the TMJ capsule. We have also summarized the differential diagnoses and sorted them into four categories. Both the clinical history and examination findings, augmented as indicated with imaging results, are needed to achieve a correct diagnosis of SC of the TMJ.

Ethics statement

The Ethics Committee of Zhejiang Provincial People's Hospital approved this study (No. 2020QT242). We have de-identified all patient details. The requirement for informed consent was waived because journal policy states that written patient consent for publication is not required if the identity of the patient cannot be ascertained in any way.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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Authors' contributions

Wenyan Zhao was responsible for drafting the manuscript and searching the literature. Wentao Zhang was in charge of performing the surgery. Fan Yang conceived the study and participated in its design and coordination. All authors read and approved the final manuscript.

Availability of data and supporting materials

Data sharing is not applicable to this article because no datasets were generated or analyzed during the current study.

CARE Checklist (2016) statement

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References

1. Lee LM, Zhu YM, Zhang DD, et al. Synovial chondromatosis of the temporomandibular joint: a clinical and arthroscopic study of 16 cases. *J Craniomaxillofac Surg* 2019; 47: 607–610.
2. Liu X, Huang Z, Zhu W, et al. Clinical and imaging findings of temporomandibular joint synovial chondromatosis: an analysis of 10 cases and literature review. *J Oral Maxillofac Surg* 2016; 74: 2159–2168.
3. Leite PCC, Tolentino EDS, Yamashita AL, et al. Surgical treatment of synovial chondromatosis in the inferior compartment of the temporomandibular joint with articular disc involvement. *J Craniofac Surg* 2018; 29: e199–e203.
4. Shah SB, Ramanojam S, Gadre PK, et al. Synovial chondromatosis of temporomandibular joint: journey through 25 decades and a case report. *J Oral Maxillofac Surg* 2011; 69: 2795–2814.
5. Ardekian L, Faquin W, Troulis MJ, et al. Synovial chondromatosis of the temporomandibular joint: report and analysis of eleven cases. *J Oral Maxillofac Surg* 2005; 63: 941–947.
6. Meng J, Guo C, Yi B, et al. Clinical and radiologic findings of synovial chondromatosis affecting the temporomandibular joint. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2010; 109: 441–448.
7. Tang B, Wang K, Wang H, et al. Radiological features of synovial chondromatosis affecting the temporomandibular joint: report of three cases. *Oral Radiol* 2019; 35: 198–204.
8. Alexiou K, Stamatakis H and Tsiklakis K. Evaluation of the severity of temporomandibular joint osteoarthritic changes related to age using cone beam computed tomography. *Dentomaxillofac Radiol* 2009; 38: 141–147.
9. Larheim TA. Current trends in temporomandibular joint imaging. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1995; 80: 555–576.
10. Liu X, Wan S, Shen P, et al. Diagnostic accuracy of synovial chondromatosis of the temporomandibular joint on magnetic resonance imaging. *PLoS One* 2019; 14: e0209739.
11. Romano A, Lo Faro C, Salzano G, et al. Synovial chondromatosis of temporomandibular joint spreading into the cranial space. *J Oral Maxillofac Surg Med Pathol* 2018; 30: 267–271.
12. August M, Faquin WC, Ferraro NF, et al. Fine-needle aspiration biopsy of intraosseous jaw lesions. *J Oral Maxillofac Surg* 1999; 57: 1282–1286.
13. Brabyn PJ, Capote A, Muñoz-Guerra MF, et al. Arthroscopic management of synovial chondromatosis of the temporomandibular joint. case series and systematic review. *J Maxillofac Oral Surg* 2018; 17: 401–409.
14. Von Lindern JJ, Theuerkauf I, Niederhagen B, et al. Synovial chondromatosis of the temporomandibular joint: clinical, diagnostic, and histomorphologic findings. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002; 94: 31–38.
15. Milgram JW. The classification of loose bodies in human joints. *Clin Orthop Relat Res* 1977: 282–291.
16. Mathew P, Tiwari RVC and Govindan NO. Temporomandibular joint synovial chondromatosis posing as diagnostic dilemma: a case report. *J Maxillofac Oral Surg* 2019; 18: 543–546.
17. Yoshida H, Tsuji K, Ishikawa H, et al. Scanning electoron microscopical analysis of the loose bodies of synovial chondoromatosis in temporomandibular joint. *Med Mol Morphol* 2020; 53: 82–85.
18. Maffia F, Vellone V, De Quarto C, et al. Synovial chondromatosis of the temporomandibular joint with glenoid fossa erosion: disk preservation for spontaneous anatomical recovery. *J Craniomaxillofac Surg* 2019; 47: 1898–1902.
19. Li Y, El Mozen LA, Cai H, et al. Transforming growth factor beta 3 involved in the pathogenesis of synovial chondromatosis of temporomandibular joint. *Sci Rep* 2015; 5: 8843.
20. Li Y, Feng Y, Cao P, et al. Role of synovium-derived fibrous cartilage in temporomandibular joint synovial chondromatosis. *J Oral Pathol Med* 2019; 48: 79–86.

21. Yoshida H, Tsuji K, Oshiro N, et al. Preliminary report of Ki-67 reactivity in synovial chondromatosis of the temporomandibular joint: an immunohistochemical study. *J Craniomaxillofac Surg* 2013; 41: 473–475.
22. Barraclough O, Wilson G and Power A. Synovial chondromatosis of the temporomandibular joint: a case report. *Ann R Coll Surg Engl* 2020; 102: e213–e215.
23. Nath P and Menon S. Synovial chondromatosis of the temporomandibular joint. *J Maxillofac Oral Surg* 2020; 19: 230–234.
24. Mupparapu M. Synovial chondromatosis of the temporomandibular joint with extension to the middle cranial fossa. *J Postgrad Med* 2005; 51: 122–124.
25. Warner BF, Luna MA and Robert Newland T. Temporomandibular joint neoplasms and pseudotumors. *Adv Anat Pathol* 2000; 7: 365–381.
26. Covani U, Orlando B, Galletti C, et al. Chondrocalcinosis of the temporomandibular joint: clinical considerations and case report. *Cranio* 2009; 27: 134–139.
27. Avinash KR, Rajagopal KV, Ramakrishnaiah RH, et al. Computed tomographic features of mandibular osteochondroma. *Dentomaxillofac Radiol* 2007; 36: 434–436.
28. Oda Y, Izumi T, Harimaya K, et al. Pigmented villonodular synovitis with chondroid metaplasia, resembling chondroblastoma of the bone: a report of three cases. *Mod Pathol* 2007; 20: 545–551.
29. Orhan K, Arslan A and Kocyigit D. Temporomandibular joint osteochondritis dissecans: case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006; 102: e41–e46.