

Synovial chondromatosis in the temporomandibular joint: case report with review of the literature

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SUMMARY

Synovial chondromatosis (SC) of the temporomandibular joint (TMJ) is a rare benign condition characterized by the formation of metaplastic cartilage in the synovium resulting in numerous attached and unattached osteocartilagenous (calcified) loose bodies within the joint. The purpose of this article is to present a case of SC of the TMJ and to discuss current diagnostic approaches, treatment options and relevant follow-up data. We present a case of SC in the TMJ that was confirmed by histopathological analysis and treated via arthrotomy, and present the typical imaging findings, including Computed Tomography (CT) and orthopantomography (OPTG) findings.

The study was financially supported by the grants IUT2-8 and ETF 6591.

Keywords: synovial chondromatosis, temporomandibular joint; joint loose bodies.

INTRODUCTION

Synovial chondromatosis (SC) of the temporomandibular joint (TMJ) is a rare pathologic lesion (benign condition) characterized by the formation of metaplastic cartilaginous nodules of the mesenchymal remnants in the synovium and inside the articular space. These nodules may present themselves as attached or unattached osteocartilagenous (calcified) loose bodies within the joint (1, 2).

According to Ginaldi (3), Ambroise Paré was the first to report this joint disease in 1558 and the involvement of the TMJ was described in 1933 by Axhausen (4).

The first histomorphologic description was provided by Jaffe in 1958, laying the foundations for histologic diagnosis of the disease (5). The first accurate scientific description of SC of the TMJ was given in 1933 by Axhausen, who described it as 'metaplastic chondrogenesis in the synovial membrane' (6).

Two forms of the SC have been recognized: primary and secondary. It may develop as a primary lesion or may arise after a trauma or another disease process such as osteoarthritis. The pathogenesis of the primary

form of SC is unknown quite uncommon. It has been proposed that the disease develops as a response to repetitive, low-grade trauma. Inflammation, parafunctions or joint overuse, as well as degeneration, have been seen as potential causative factors for the secondary form (7, 8). SC is observed in 17% of patients treated arthroscopically due to TMJ osteoarthritis (9). This situation often is causing dysfunction of the TMJ, variable enlargement of the joint capsule and suggests a broad differential diagnosis that spans degenerative joint disease and cartilaginous neoplasms including chondrosarcoma (10, 11).

SC usually affects large synovial joints, such as the elbow, hip, wrist and knee, occurring twice as often among men compared to women (12).

The TMJ to be rarely affected by SC, having the prediction to occur mostly in the upper compartments (13, 14), in the lower compartment it is extremely rare (15, 16).

Blankestijn et al. described 3 stages in the genesis of SC of the TMJ (17). The primary stage involves metaplasia in the synovial membrane without loose bodies. In the second phase, progressive metaplasia leads to detachment of loose bodies. These contain active chondrocytes and are partially surrounded by a synovial membrane. In the final phase there is no longer any metaplastic activity in the region of the synovial membrane. This allows the loose bodies to degenerate and calcify.

A review of the literature up to the year 2000 revealed at least 79 cases of SC that have involved

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the TMJ (18). Since then several additional cases of SC of the TMJ have been reported.

Clinical manifestations, radiological images as orthopantomography (OPTG), Computed Tomography (CT), Magnetic Resonance Imaging (MRI) and arthroscopy are used in diagnosing SC. A pathohistological analysis is mandatory to confirm the disease (7, 19).

The clinical features of SC in the TMJ are pain, swelling, limitation of the movements of the jaw and crepitation or clicking sound when opening the mouth, malocclusion, vertigo, and tinnitus (20). The presence of cranial nerve dysfunction indicates that the disease has reached an advanced stage. SC may extend from TMJ to the surrounding tissues: parotid gland, middle ear, intracranial space (21). The first case of this type was reported in 1977 (22).

The purpose of this article is to present a case of SC of the TMJ and to discuss current diagnostic approaches, treatment options and relevant follow-up data. The data presented in the literature regarding SC in the TMJ will also be reviewed and analysed.

CASE REPORT

A 45 year old woman was referred in June 2011 with a chief complaint of hearing disturbances in the left TMJ and preauricular tenderness, a maximal mouth opening of 38 mm with a slight deviation to the left, and normal occlusion, and a clicking sound in the left joint during mouth opening.

In additional TMJ pathology the patient had a series of involvement other joint disease. There was a history of bilateral coxarthrosis, with insertion of left hip prosthesis, bilateral gonarthrosis, with right knee arthroscopy and recently was diagnosed polyarthrosis. 20 years ago the patient had myocardial infarction. There was no history of any trauma.

Initial imaging studies included OPTG (CRAEX 3, Soredex orion corporation LTD, Finland) and CT scans (SOMATOM AR HP Spiral, Siemens, Erlangen, Germany). OPTG revealed radio-opacity in the left TMJ region showing large calcified masses in the left TMJ space (Figure 1). Axial and coronal CT images showed distinct nodules within an extremely expanded upper joint compartment and also revealed granular masses in the joint space surrounding the left condylar head

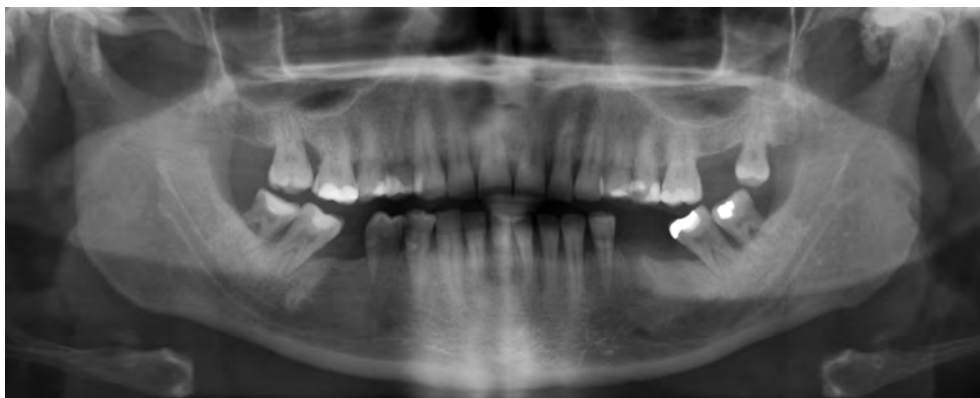


Fig. 1. Ortopantomograph. OPTG presenting a picture of SC in the left TMJ. Radio-opacity in the left TMJ region showing large calcified masses.

(Figures 2, 3). No bone changes were found. The right TMJ was normal.

An arthrotomy was performed under nasotracheal general anaesthesia in order to explore the left TMJ. A preauricular incision was made to expose the left articular capsule. The capsule was incised laterally and the upper and the lower compartments of the joint were explored. Numerous opalescent glistening loose bodies were found in the superior joint compartment, which also exhibited hyperaemia of the synovial membrane (Figure 4). Irregular cartilaginous loose granules of various sizes were found and removed from the upper compartment of the TMJ. The nodules were pearly white, of varying shapes, ranging in diameter from 3 to 10 mm (Figure 5). The compartment was explored, irrigated with saline solution.

In this case loose bodies were present in the upper space and a discopexia of the anterior disc was performed. The wound was closed in layers.

The patient was discharged from hospital with anti-inflammatory and anti-analgesic medications (Penicillin, Paracetamol). In the postoperative period the patient engaged in active articular physiotherapy.

The diagnosis was confirmed by histological investigation. Histopathological findings showed chondrometaplasia of the synovial membrane. The nodules were of cellular hyaline cartilage covered by a fine fibrous layer, and sometimes by synovial lining cells without focal osteoid formation (Figures 6, 7). The absence of invasions into the surrounding tissues was important to exclude the possibility of malignancy. No radiographic evidence of recurrence or residual disease was detected in the operated-on joint after 6 months.

DISCUSSION

SC is a proliferative disorders of the synovium that is associated with the formation of metaplastic cartilaginous or osteocartilaginous nodules in the synovial membrane which eventually detach as loose bodies.

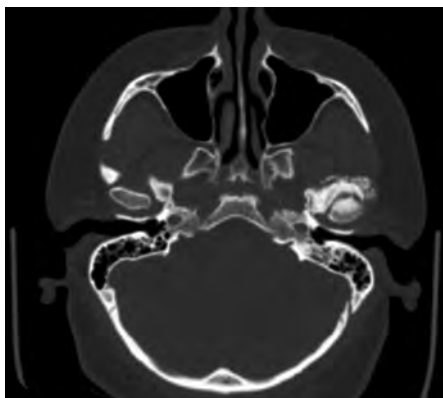


Fig. 2. Computed Tomography. Axial CT view showing distinct nodules within an extremely expanded upper joint compartment revealing granular masses in the joint space, surrounding the left condylar head. The right TMJ is normal.

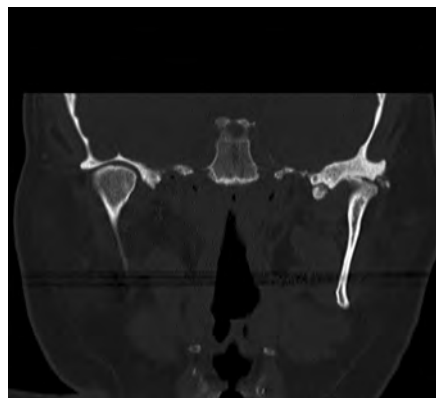


Fig. 3. Computed Tomography. Coronal view of the CT. Radioopacity of the left TMJ compartment.



Fig. 4. Arthrotomy. The TMJ space is opened by arthrotomy. The capsule is incised, numerous opalescent glistening loose nodules are visible.



Fig. 5. The nodules. Pearly white nodules of varying shape removed in the course of surgery, ranging in diameter from 3 to 10 mm.



Fig. 6. Histological investigation. A cartilaginous nodule with clustered chondrocytes, lined by connective tissue resembling the synovium. H & E $\times 40$.

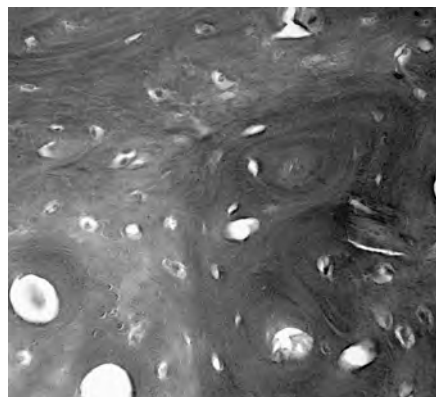


Fig. 7. Histological investigation. Magnified view of a tissue section showing details of chondrocytes. H & E $\times 400$.

According to Fujita (23) TGF- β and tenascin may be implicated in chondrogenesis in all phases of SC. The case presented here is unusual because of its obvious history of systemic disease (rheumatoid arthritis) with involvement of the TMJ.

Our findings suggest that synovial chondromatosis of the TMJ affects only the synovial lining of the upper compartment of the joint. It is likely that the loose bodies in the inferior joint space originated from the upper compartment of the joint, penetrating into the inferior compartment after the perforation of the articular disc (13). As long as the synovial membrane

is present and receives some pressure, SC can develop in the inferior compartment of the TMJ. The area of the synovial membrane in the lower compartment is smaller than in the upper compartment, and its ability to produce loose bodies must therefore be inferior to that of the upper compartment (24).

Most of the literature available on this topic originates in Asian countries suggesting an ethnic predisposition or underreporting and/or underdiagnosis in the Western countries. Systematic reviews of the literature on SC of the TMJ showed that the mean age of presentation is between the 4th and the 5th decade, the presenta-

tion tends to be unilateral and with a preponderance of female cases (20, 21, 25). Bilateral involvement have been reported only in three cases (26-28).

The histological appearance is that of a benign chronic inflammation of the synovium varying in severity and with metaplastic activity. In 1977 Milgram described a 3-phase course of synovial chondromatosis in the limb joints. According to this description, the first stage involves metaplastic changes in the synovial membrane without the presence of detached particles, the second stage shows metaplasia of the synovial membrane with the presence of detached particles and the third stage presents with detached particles only, which may vary in diameter from less than 1 mm to greater than 10 mm (29).

In our experience and the information in the literature the correlation of histologic and radiologic findings was considered most consistent with SC.

The symptoms of SC are non-specific and diagnosis is based on imaging and histological investigation. The main imaging features of SC in the TMJ are as follows: widening of the joint space, soft tissue swelling, multiple calcified loose bodies, irregular surfaces of joints and sclerosis or hyperostosis of the glenoid fossa and the mandibular condyle. Coronal CT images are particularly good for detecting loose bodies in the joint spaces (4, 7). These findings are non-specific, most of them may also be observed in osteoarthritis of the TMJ, which is the most common pathology for this joint, in intracapsular fractures or inflammatory arthritis and in the more aggressive pathologies such as chondrosarcoma (30).

Arthroscopy is a reliable diagnostic tool for SC because of the direct visualization of the joint space and allows to diagnose the disease at an early stage (31, 32).

It is suggested that treatment of SC should be selected in accordance with the stage of the disease from among options ranging from open joint surgery to remove loose bodies and the affected synovium to condylectomy, discectomy, and reconstruction with a costochondral graft and pedicled deep temporal fascial fat flap (33).

Sanroman et al. found that most patients with SC can be treated by arthroscopy removal of the loose bodies, with coagulation of the affected synovium by means of conventional bipolar electrocautery or radiofrequency devices (8). Arthroscopy has been successful in cases where the mass lesion is confined

to a single joint compartment, does not exhibit extra-articular extension and the particle size is less than 3 mm (9, 32, 34, 35).

The literature did not provide any useful information on the correlation between the stage of the disease and the type of surgical intervention. It seems that the surgeon's experience is the main determinant of the additional procedures to be performed as well as loose body removal (20).

It is important to differentiate SC from other diseases, like calcium pyrophosphate dihydrate crystal deposition disease of the TMJ which is also termed tophaceous pseudogout or chondrocalcinosis (36).

Malignant transformation of SC of the TMJ appears to be very uncommon. Some authors reports a relative 5% malignancy risk of SC in large joints (18). In the English literature only three cases of malignant transformation of SC involving the TMJ have been described (37-39).

In summary, SC is a benign disease with a broad differential diagnosis, which does not respond to non-surgical treatment and does not show spontaneous resolution. Early diagnosis is very important for selecting appropriate treatment and for a better prognosis.

CONCLUSION

As symptoms are nonspecific, the diagnosis of SC is based mainly on imaging and histological investigation. Open arthrotomy would be required if particle size exceeds 3 mm. Removal of all the involved synovium, and cartilaginous bodies is important for adequate treatment.

ACKNOWLEDGMENTS

The study was financially supported by the grants IUT2-8 and ETF 6591. The authors would like to thank Dr. Triin Erm from the Department of Pathology of the Tartu University Hospital for the pathohistological investigation and interpretation and Mr. Andrus Aavik from the Department of Radiology of the Tartu University Hospital.

STATEMENT OF CONFLICT OF INTEREST

The authors have no financial disclosures to report.

REFERENCES

1. Ardekian L, Faquin W, Troulis MJ, Kaban LB, August M. Synovial chondromatosis of the temporomandibular joint: report and analysis of eleven cases. *J Oral Maxillofac Surg* 2005;63:941-47.
2. Lieger O, Zix J, Stauffer-Brauch EJ, Iizuka T. Synovial chondromatosis of the temporomandibular joint with cranial extension: a case report and literature review. *J Oral Maxillofac Surg* 2007;65:2073-80.

3. Ginaldi S. Computed tomography feature of synovial osteochondromatosis. *Skeletal Radiol* 1980;5:219-22.
4. Wong WC, Cheng PW, Chan FL. MRI appearance of synovial chondromatosis in the temporomandibular joint. *Clin Radiol* 2001;56:773-74.
5. Jaffe HL. Tumours and tumorous conditions of the bones and joints. Kimpton, London; 1958.
6. Axhausen G. Pathologie und Therapie des Kiefergelenks. *Fortschr Zahnheilk*;1993.
7. Martín-Granizo R, Sánchez JJ, Jorquera M, Ortega L. Synovial chondromatosis of the temporomandibular joint: a clinical, radiological and histological study. *Med Oral Patol Oral Cir Bucal* 2005;10:272-76.
8. Sanromán JF, López AC, Badiola IA, Ferro MF, DE Sánchez AL. Indications of arthroscopy in the treatment of synovial chondromatosis of the temporomandibular joint: report of 5 new cases. *J Oral Maxillofac Surg* 2008;66:1694-99.
9. Leibur E, Jagur O, Mürsepp P, Veede L, Voog-Oras Ü. Long-term evaluation of arthroscopic surgery with lysis and lavage of temporomandibular joint disorders. *J Craniomaxillofac Surg* 2010;38:615-20.
10. Coleman H, Chandraratnam E, Morgan G, Gomes L, Bonar F. Synovial chondrosarcoma arising in synovial chondromatosis of the temporomandibular joint. *Head Neck Pathol* 2013;7:304-09.
11. Sink J, Bell B, 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:e269-74.
12. Koyama J, Ito J, Hayashi T, 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;22:1203-06.
13. Holmlund AB, Eriksson L, Reinholt FP. Synovial chondromatosis of the temporomandibular joint: clinical, surgical and histological aspects. *Int J Oral Maxillofac Surg* 2003;32:143-47.
14. Lim SW, Jeon SJ, Choi SS, Choi KH. Synovial chondromatosis in the temporomandibular joint: a case with typical imaging features and pathological findings. *Br J Radiol* 2011; 84:e213-16.
15. Chen MJ, Yang C, Zhang XH, Qiu YT. Synovial chondromatosis originally arising in the lower compartment of temporomandibular joint: a case report and literature review. *J Craniomaxillofac Surg* 2011;39:459-62.
16. Matsumoto K, Sato T, Iwanari S, Kameoka S, Oki H, Komiyama K, et al: The use of arthrography in the diagnosis of temporomandibular joint synovial chondromatosis. *Dentomaxillofac Radiol* 2013;42:15388284.
17. Blankestijn J, Pander AK, Vermey A, Scherpbier AJ. Synovial chondromatosis of the temporo-mandibular joint. Report of three cases and review of the literature. *Cancer* 1985;55:479-85.
18. Warner BF, Luna MA, Robert Newland T. Temporomandibular joint neoplasms and pseudotumors. *Adv Anat Pathol* 2000;7:365-81.
19. Wang P, Tian Z, Yang J, Yu Q. Synovial chondromatosis of the temporomandibular joint: MRI findings with pathological comparison. *Dentomaxillofac Radiol* 2012;41:110-16.
20. Guarda-Nardini L, Piccotti F, Ferronato G, Manfredini D. Synovial chondromatosis of the temporomandibular joint: a case description with systematic literature review. *Int J Oral Maxillofac Surg* 2010;39:745-55.
21. Kahraman AS, Kahraman B, Dogan M, Firat C, Samdanci E, Celik T. Synovial chondromatosis of the temporomandibular joint: radiologic and histopathologic findings. *J Craniofac Surg* 2012;23:1211-13.
22. Noyek AM, Holgate RC, Fireman SM, Rosen P, Pritzker KP. The radiological findings in synovial chondromatosis (chondrometaplasia) of the temporomandibular joint. *J Otolaryngol* 1977;6:45-48.
23. Fujita S, Yoshida H, Tojyo I, Wada T, Murakami K, Iizuka T. Synovial chondromatosis of the temporomandibular joint: clinical and immunohistopathological considerations. *Br J Oral Maxillofac Surg* 2004;42:259-60.
24. Sato J, Notani KI, Goto J, Shindoh M, Kitagawa Y. Synovial chondromatosis of the temporomandibular joint accompanied by loose bodies in both the superior and inferior joint compartments: case report. *Int J Oral Maxillofac Surg* 2010;39:86-88.
25. Cai XY, Yang C, Chen MJ, Jiang B, Zhou Q, Jin JM, et al. Arthroscopic management for synovial chondromatosis of the temporomandibular joint: a retrospective review of 33 cases. *J Oral Maxillofac Surg* 2012;70:2106-13.
26. Keogh CF, Torreggiani WC, Munk PL. Bilateral synovial chondromatosis of the temporomandibular joint. *Clin Radiol* 2002;57:862.
27. Peng LW, Yan DM, Wang YG, Li YD. Synovial chondromatosis of the temporomandibular joint: a case report with bilateral occurrence. *J Oral Maxillofac Surg* 2009;67:893-95.
28. Guijarro-Martínez R, Puche Torres M, Marqués Mateo M, Solís García I, Miragall Alba L, Iglesias Gimilio ME, et al: Bilateral synovial chondromatosis of the temporomandibular joint. *J Craniomaxillofac Surg* 2011;39:261-65.
29. Milgram JW. The classification of loose bodies in human joints. *Clin Orthop Relat Res* 1977;124:282-91.
30. Balliu E, Medina V, Vilanova JC, Peláez I, Puig J, Trull JM, et al. Synovial chondromatosis of the temporomandibular joint: CT and MRI findings. *Dentomaxillofac Radiol* 2007;36:55-8.
31. Cai XY, Yang C, Chen MJ, Jiang B. Arthroscopically guided removal of large solitary synovial chondromatosis from the temporomandibular joint. *Int J Oral Maxillofac Surg* 2010;39:1236-39.
32. Leibur E, Jagur O, Voog-Oras Ü. Temporomandibular joint arthroscopy. In: Jason L. Dragoo. *Modern Arthroscopy*. INTECH-Open Access Publisher; 2011. p.3-26.
33. Chen MJ, Yang C, Cai XY, Jiang B, Qiu YT, Zhang XH. Synovial chondromatosis in the inferior compartment of the temporomandibular joint: different stages with different treatments. *J Oral Maxillofac Surg* 2012;70:e32-38.
34. McCain JP, de la Rua H. Arthroscopic observation and treatment of synovial chondromatosis of the temporomandibular joint. Report of a case and review of the literature. *Int J Oral Maxillofac Surg* 1989;18:233-36.
35. Leibur E, Jagur O, Voog-Oras Ü. Temporomandibular Joint. Arthroscopy vs Arthrotomy. In: *Vaibhav Bagaria. Regional Arthroscopy*. INTECH- Open Access Publisher; 2013. p.61-95.
36. Nakagawa Y, Ishibashi K, Kobayashi K, Westesson PL. Calcium pyrophosphate deposition disease in the temporomandibular joint: report of two cases. *J Oral Maxillofac Surg* 1999;57:1357-63.
37. Merrill RG, Yih WY, Shamloo J. Synovial chondrosarcoma of the temporomandibular joint: a case report. *J Oral Maxillofac Surg* 1997;55:1312-16.
38. Ichikawa T, Miyauchi M, Nikai H, Yoshiga K. Synovial chondrosarcoma arising in the temporomandibular joint. *J Oral Maxillofac Surg* 1998;56:890-94.
39. Yokota N, Inenaga C, Tokuyama T, Nishizawa S, Miura K, Namba H. Synovial chondromatosis of the temporomandibular joint with intracranial extension. *Neurol Med Chir* 2008;48:266-70.

Received: 14 03 2014

Accepted for publishing: 28 09 2015