

Intra- and extracapsular synovial chondromatosis of the temporomandibular joint: Rare case and review of the literature

SAGE Open Medical Case Reports
Volume 6: 1–5
© The Author(s) 2018
Reprints and permissions:
sagepub.co.uk/journalsPermissions.nav
DOI: 10.1177/2050313X18775307
journals.sagepub.com/home/sco



Henrik Holtmann¹ , Thomas Böttinger¹, Norbert R Kübler¹,
Daman D Singh¹, Christoph K Sproll¹, Karline Sander¹,
Renan Langie², Felix Schrader¹, and Julian Lommen¹

Abstract

Synovial chondromatosis is a benign disease which most commonly appears in large joints and only rarely affects the temporomandibular joint. The diagnosis is quite difficult due to the fact that a large swelling in the preauricular area and the radiographic findings may be misdiagnosed as other benign or malignant diseases. We report an unusual case of intra- and extracapsular chondromatosis of 25 osteochondral loose bodies in the right temporomandibular joint.

Keywords

Otolaryngology, radiology, surgery, synovial chondromatosis, temporomandibular joint, head and neck

Date received: 5 February 2018; accepted: 16 April 2018

Introduction

Synovial chondromatosis (SC) is a rare benign lesion which includes osteochondral loose bodies and normally occurs in large joints such as the knee, elbow or hip. The temporomandibular joint (TMJ) is the rarest location of this disease.¹ The current literature review reveals only around 200 previous cases of SC affecting the TMJ.^{2,3} We report an unusual case of intra- and extracapsular chondromatosis of 25 osteochondral loose bodies in the right TMJ.

Case report

A 63-year-old woman was referred by her general medical practitioner to our Department for Oral and Maxillofacial Surgery. The patient noticed an increasing painful swelling in the right preauricular region during the past 2 years. The dental situation included a prosthetic insufficient treated status and a strong abrasion of the teeth most likely due to a supposed craniomandibular dysfunction. There was no previous history of trauma. One year before there had been an excision of a malignant melanoma (pT1a, cN0, cM0), in the skin of the left knee. The patient suffered from no severe disease besides arterial hypertension and rhinoconjunctivitis allergica.

Performing an extraoral examination, there was a marked facial asymmetry with deviation of the chin to the left side and a remarkable bony hard swelling in the right condylar

region. Mouth opening was possible with no pronounced clicking or crepitus.

The patient wore partial dentures with 5 own remaining teeth in the upper jaw and 9 remaining teeth in the lower jaw. The occlusion was an Angle Class I with correct denticulation but seemed to be a skeletal mandibular protrusion in the side profile clinically because of a prominent chin. The condyles itself looked similar in size and form (Figure 1).

In the first sonography, unsharp circumscribed possibly calcified lesions could be seen in the cranial of the right caput mandibulae. There was no increased vascularisation to be found. Beside this, the TMJs on both sides were in a symmetric degenerative status.

The full extent of these lesions on the right side was defined more precisely by a computed tomography (CT) scan

¹Department of Oral and Maxillofacial Surgery, University Hospital Düsseldorf, Düsseldorf, Germany

²Santa Casa de Misericórdia de Porto Alegre (SCMPA), Avenida Independência, Porto Alegre, Brazil

Corresponding Author:

Henrik Holtmann, Department of Oral and Maxillofacial Surgery, University Hospital Düsseldorf, Moorenstraße 5, Building 11.52, 40225 Düsseldorf, Germany.
Email: Henrik.Holtmann@med.uni-duesseldorf.de





Figure 1. Praeoperative orthopantomography of a 63-year-old patient with synovial chondromatosis of the right temporomandibular joint.

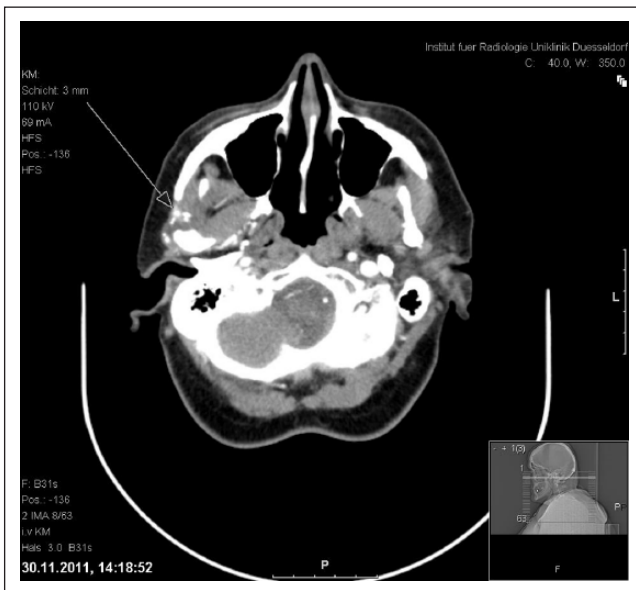


Figure 2. Praeoperative CT scan of the lesion in the right temporomandibular joint.

(Figure 2). Diverse loose calcified bodies which appeared to be within and outside of the right capsule could be recognized in form of a remarkable saculation to the medial direction.

In a separate performed magnetic resonance imaging (MRI), lesions could furthermore be described as a cystical diagnostic finding (Figures 3–5). The suspected radiological differential diagnoses after CT and MRI was therefore an (ectopic) adenoma of the right parotid gland or a saculation of the joint capsule with calcifications. An evidence for a malignant lesion could not be pointed out in CT and MRI. Therefore, no further medical imaging was performed.

Afterwards, a sonographically supported aspiration biopsy was performed to verify the final diagnosis: The cytological survey did not show any correlation to a malignant melanoma metastasis. The cells were suspected to be of benign tumor origins and most likely to stand in correlation to a pleomorph adenoma of the right parotid gland.

In view of these findings and the progressive swelling in the right TMJ, an open operative exploration was performed. The access was obtained by a right preauricular approach in order to gain enough exposure of the numerous small particles in the temporomandibular region. Twenty five loose bodies of different size were retrieved from the synovial space and interior of the capsule (Figure 6).

Closure was performed in multi-layers and a mini suction drain was inserted. The macroscopic examination of the findings resulted in biggest pieces of 14 mm x 6 mm and all bodies seemed to be of chondroid origin. This was verified by the microscopic examination which revealed all bodies to be of calcified chondral origin with regressive changes with characteristics most likely of a SC (Figure 7).

Discussion

The literature about SC has its origin in a first description by Ambroise Pare in 1558, followed by Lannec in 1813.² It was reported as a monoarticular, metaplastic change of the synovium with final formation of hyaline cartilage nodules in a joint, in particular the knees (35%) and the elbows (22%).⁴

With only 3% of all reported cases, SC is uncommon in the TMJ.^{3,5} This first discovery of the SC in the TMJ was made by Axhausen⁶ in 1933. Since then, only around 200 cases of SC affecting the TMJ were reported in the literature. In general, women are affected more frequently by SC than men.^{2,7,8}

Milgram classified this disease histopathologically into 3 types: (1) a lesion without detached bodies, (2) a lesion with synovial metaplasia and presence of loose bodies, and (3) a lesion with loose bodies and intact synovium.⁹

In our case, there was an expanded joint space surrounded by 25 loose calcified bodies. No bony destruction of the condyle, glenoid fossa or temporal bone could be found. According to histological and CT findings, our case could be classified as type 2 of Milgram's classification.

The usual symptoms of SC include painful preauricular swelling, decreased range of motion and crepitation. Often, it is misdiagnosed as internal derangement of the TMJ because of its low incidence and atypical clinical features.⁷

Diagnostics of the SC with an identification of the calcified cartilaginous bodies and bony erosions can be done by CT and MRI. Initial radiographic findings are mostly not visible with panoramic radiography.^{10,11}

The advantage of the MRI is the detection of cartilaginous nodules at an early stage even if they are not calcified. Also the evaluation of the disk, the intra-articular lesion and the extra-articular extension is possible.¹²

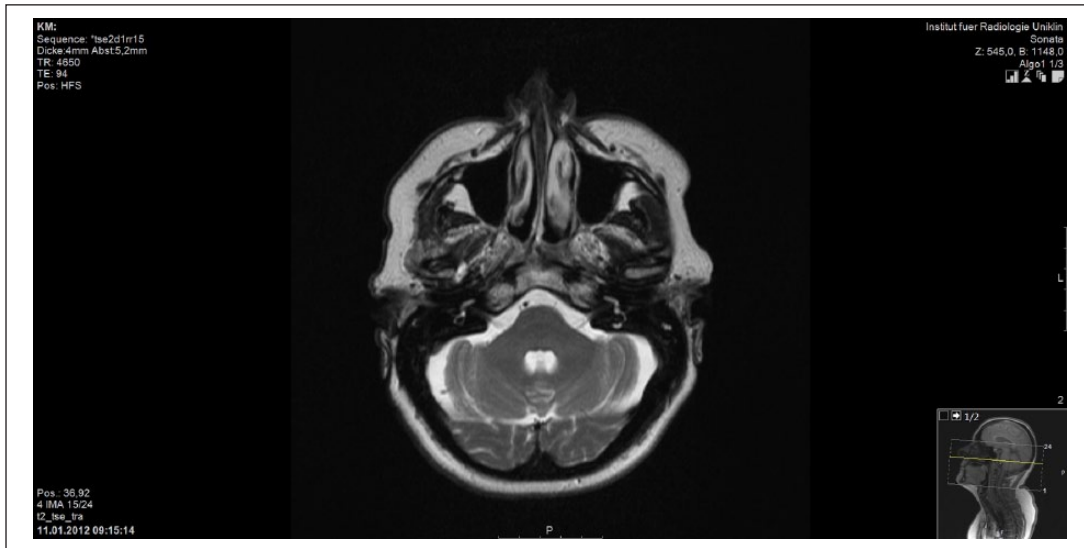


Figure 3. Preoperative MRI. Axial T2-weight slide. Praeliminary lesion surrounding the right temporomandibular joint.

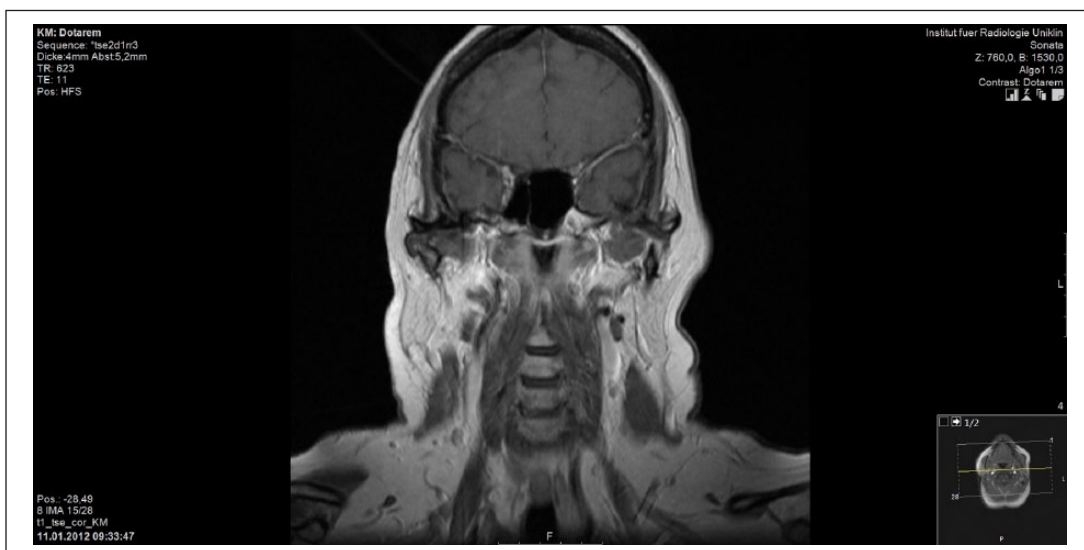


Figure 4. Preoperative MRI: coronal slide, contrast-enhanced T1-weight—cranio-lateral surrounding of the TMJ by the lesion.

In our case, the numerous calcified bodies could also be clearly detected in the CT. Nevertheless, CT and MRI do not always lead to a correct diagnosis because of possible minimal calcification of the loose bodies and their low intensity.

There have also been several reported cases in which CT and MRI were not effective in diagnosis but retroscopy was.^{13,14} Therefore, and to rule out malignant lesions sometimes, positron emission tomography (PET)-CT, gadolinium- or technetium-scintigraphy is recommended in the actual literature.³

The differential diagnosis of calcified loose bodies found within TMJ includes SC, chondrosarcoma and other benign inflammatory diseases such as osteoarthritis, rheumatoid

arthritis and avascular necrosis.¹⁵ SC and synovial chondrosarcoma have many similarities in clinical, radiographic and histological features.

By a description of Bertoni et al.¹⁶ the following histological criteria for the diagnosis of SC in differentiation from synovial chondrosarcoma can be seen: loss of clustering growth pattern, myxoid changes in the matrix, presence of necrosis, hypercellularity and permeation of the trabecular bone. In our case there was no histological evidence of necrosis but the chondrocytes showed clustering of the periphery with cellular atypia like enlarged nuclei and binucleated cells and focal increased cellularity (Figure 5).

The final therapy, however, is open arthroscopy according to McCain and De la Rúa¹⁷ if loose bodies are larger than

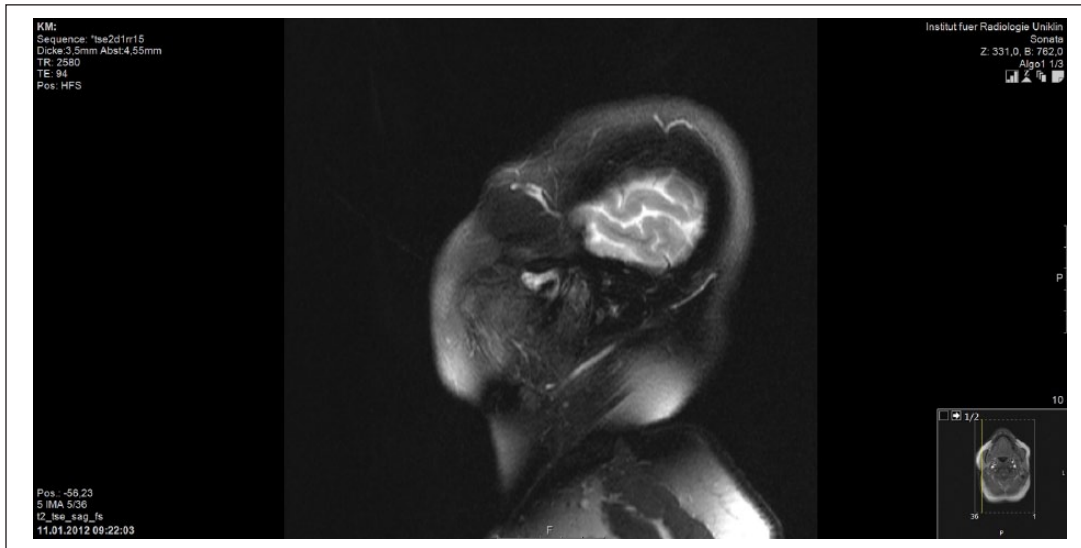


Figure 5. Preoperative MRI: Sagittal T2-weight-slide of the right TMJ showing the high intensity lesion.



Figure 6. Twenty five loose bodies of different size retrieved from the synovial space and the inner TMJ-capsule.

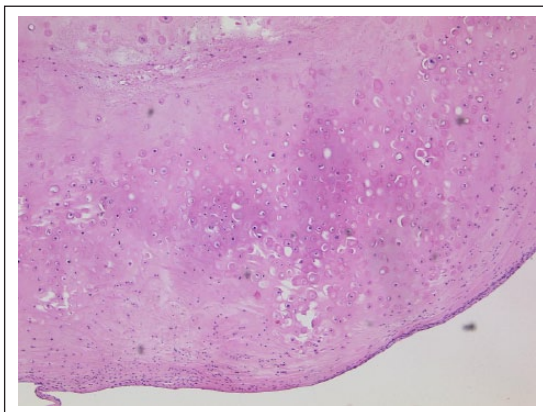


Figure 7. Calcified chondral bodies with regressive changes most likely to correlation to a synovial chondromatosis.

3 mm. Undoubtedly, thoroughly complete removal of the loose bodies provided by the surgeon's experience is decisive for the final result.

Recurrence is likely if cartilaginous foci or hyperplastic synovium are incompletely removed but unlikely even after sparing extracapsular particles, provided total synectomy and disectomy are performed.^{3,18} So once diagnosed, removal of loose bodies and the synovium with or without condylectomy is the recommended and accepted treatment to prevent recurrence because the condition does not undergo spontaneous resolution. Malignant transformation of SC to chondrosarcoma has very rarely been reported in the knee joint¹⁹ but may also be possible in the TMJ.

Almost all cases of SC of the TMJ reported monoarticular occurrence. Some authors hold the opinion that SC of the TMJ has a prediction for the right side. Our reported case is in accordance to these descriptions. Nevertheless, this is the focus of discussion. The early stage of SC of the TMJ is manifested not necessarily by abnormality or spreaded calcification of the loose bodies but by smaller processes that are not obvious in CT scans. Therefore, it is understandable why SC of the TMJ so very often presents so-called unilateral prediction. Especially as we know cases with bilateral occurrence.²⁰

So it seems to be elementary not to overlook checking the contralateral TMJ region and even in case of no abnormality to be found to carry out a long-term follow-up.

Conclusion

The diagnosis to a SC is challenging due to the fact that a large swelling in the preauricular area and the radiographic findings may be misdiagnosed as other benign or malignant diseases and the fact a SC is a rare finding in that area. The

open surgical removal of loose bodies and the synovium with or without condylectomy is the recommended and accepted treatment which leads to very low recurrence rates. A malignant transformation to a chondrosarcoma has very rarely been reported in the knee joint but may also be possible in the TMJ.

Declaration of conflicting interests

The author(s) declared the following potential conflicts of interest with respect to the research, authorship, and/or publication of this article: All authors disclaim any financial or non-financial interests or commercial associations that might pose or create a conflict of interest with information presented in this manuscript.

Ethics approval

Our institution does not require ethical approval for reporting individual cases or case series.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

ORCID iD

Henrik Holtmann  <https://orcid.org/0000-0003-1477-6400>

References

1. Kahraman AS, Kahraman B, Dogan M, et al. Synovial chondromatosis of the temporomandibular joint: radiologic and histopathologic findings. *J Craniofac Surg* 2012; 23(4): p1211–1123.
2. Shah SB, Ramanojam S, Gadre PK, et al. Synovial chondromatosis of temporomandibular joint: journey through 25 decades and a case report. *J Oral Maxil Surg* 2011; 69(11): 2795–2814.
3. Leite PCC, Tolentino ES, 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: 199–203.
4. Balasundaram A, Geist JR, Gordon SC, et al. Radiographic diagnosis of synovial chondromatosis of the temporomandibular joint: a case report. *J Can Dent Assoc* 2009; 75(10): 711–774.
5. Hohlweg-Majert B, Metzger MC, Böhm J, et al. Advanced imaging findings and computer-assisted surgery of suspected synovial chondromatosis in the temporomandibular joint. *J Magn Reson Imaging* 2008; 28(5): 1251–1257.
6. Axhausen. Pathologie und thearpiedeskiefergelenkes. *Fortschr Zahnheil* 1933; 9: 171–186.
7. Ardekian L, Faquin W, Troulis MJ, et al. Synovial chondromatosis of the temporomandibular joint: report and analysis of eleven cases. *J Oral Maxil Surg* 2005; 63(7): 941–997.
8. Delantoni A, Spyropoulou E, Chatziagiannis J, et al. Sole radiographic expression of rheumatoid arthritis in the temporomandibular joints: a case report. *Oral Surg Oral Med O* 2006; 102(4): e37–40.
9. Milgram JW. The classification of loose bodies in human joints. *Clin Orthop Relat R* 1977(124): 282–291.
10. Uotila E. The temporomandibular joint in adult rheumatoid arthritis: a clinical and roentgenologic study. *Acta Odontol Scand* 1964; 22(Suppl. 39): 1–91.
11. Franks AS. Temporomandibular joint in adult rheumatoid arthritis: a comparative evaluation of 100 cases. *Ann Rheum Dis* 1969; 28(2): p139–145.
12. Testaverde L, Perrone A, Caporali L, et al. CT and MR findings in synovial chondromatosis of the temporo-mandibular joint: our experience and review of literature. *Eur J Radiol* 2011; 78(3): 414–448.
13. Cai XY, Yang C, Chen MJ, et al. Simultaneous pigmented villonodular synovitis and synovial chondromatosis of the temporomandibular joint: case report. *Int J Oral Maxillof Surg* 2009; 38(11): 1215–1218.
14. Wise DP and Ruskin JD. Arthroscopic diagnosis and treatment of temporomandibular joint synovial chondromatosis: report of a case. *J Oral Maxil Surg* 1994; 52(1): 90–93.
15. Nussenbaum B, Roland PS, Gilcrease MZ, et al. Extra-articular synovial chondromatosis of the temporomandibular joint: pitfalls in diagnosis. *Arch Otolaryngol Head Neck Surg* 1999; 125(12): 1394–1397.
16. Bertoni F, Unni KK, Beabout JW, et al. Chondrosarcomas of the synovium. *Cancer* 1991; 67(1): 155–162.
17. McCain dela Rua JPH. Arthroscopic observation and treatment of synovial chondromatosis of the temporomandibular joint: report of a case and review of the literature. *Int J Oral Maxillof Surg* 1989; 18(4): 233–236.
18. Demeulemeester LJ, Bossuyt M, Casselman J, et al. Synovial chondromatosis of the temporomandibular joint. *Int J Oral Maxillof Surg* 1988; 17(3): 181–182.
19. Mullins F, Berard CW and Eisenberg SH. Chondrosarcoma following synovial chondromatosis. *Cancer* 1965; 18: 1180–1188.
20. Peng LW, Yan DM, Wang YG, et al. Synovial chondromatosis of the temporomandibular joint: a case report with bilateral occurrence. *J Oral Maxil Surg* 2009; 67(4): 893–895.