

Synovial Osteochondromatosis of Temporomandibular Joint – Diagnosis and Management

SUMMARY

Introduction: Synovial osteochondromatosis is a rare disease related to pathological changes involving joints' synovium. In 60% it concerns knee joint. In temporomandibular joint this diagnosis has been recorded in only few international medical centres. **Case report:** In the study, a case of 31 years old woman who reported to the Maxillofacial Surgery Ward of 10th Military Research Hospital and Polyclinic in Bydgoszcz, is presented. Main symptoms were tumor in the left temporomandibular joint area occurring for past 7 years, followed by swelling and pain. In the process of diagnostics and treatment multiple radiological imaging were involved. Also a surgical procedure was performed which included open revision and articular cavity lavage of the left temporomandibular joint. **Conclusions:** Treatment results, short-term, long-term observation and retrospective analysis show the way and dynamics of described condition development, benefits from surgical intervention as well as risk of recurrence.

Keywords: Osteochondromatosis, Synovial Chondromatosis, Synovial Osteochondromatosis, SOC

Jakub Wójcicki, Maciej Rozwadowski, Natasza Czajkowska, Marcin Jaskólski

Maxillofacial Surgery Ward of 10th Military Research Hospital and Polyclinic, Bydgoszcz, Poland

CASE REPORT (CR)

Balk J Dent Med, 2023;124-127

Introduction

Synovial osteochondromatosis (SOC) is a rare, benign synovial membrane mataplasia. The membrane builds the inner layer of articular capsule, its function is to facilitate joint movement and to prevent articular surfaces from excessive attrition which may lead to their serious damage. The symptoms of the disease are non-specific and the main one is joints disfunction and pain¹. SOC is mainly located in knee joints (about 50-65% of cases)² usually unilaterally. Temporomandibular joint location was noted in a few articles from all over the world^{3,4}. Sex predilection may seem interesting as SOC located in upper and lower limb joint is characteristic for male patients whereas temporomandibular location is most frequently noted in female patients⁵.

There are two types of synovial osteochondromatosis - primary and secondary, with different etiology, symptoms and course. The secondary type is developed due to a trauma which causes damage of joint structures,

especially the synovial membrane. Primary one's etiology is still not well known but it has a more dynamic course and a higher risk of recurrence^{4,6}.

Synovial fluid composition is fixed and does not change significantly, it mainly consists of hyaluronic acid of high viscosity and sparse macrophages⁷. When SOC occurs cartilaginous bodies are being produced inside of synovial cavity immersed in the synovial fluid. They cause impairment or even total impossibility of articular surfaces' movement. In primary form of SOC, the cartilaginous bodies are irregularly shaped with sharp edges whilst those in secondary form are more smooth and rounded. If not calcified those bodies may be invisible for classical radiographic imaging such as orthopantomogram what may be the main cause of proper diagnosis delay.

The aim of this paper was to present a case of 31 years old woman who reported to the Maxillofacial Surgery Ward of 10th Military Research Hospital and Polyclinic in Bydgoszcz.

Case report

31 y.o. female patient reported to the Maxillofacial Surgery Ward of 10th Military Reserach Hospital and Policlynic in Bydgoszcz because of oedema (Figure 1) and pain of preauricular area with mandible movement disturbance (Figure 2) which presented for the first time about 7 years ago. After dental checkup and orthopantomographic imaging, the dentist has diagnosed lack of any occlusal or teeth disorder. Next OPG was taken 12 months later (Figure 3). Both radiographs did not reveal any pathological process which could have been a cause of patient's complaints.



Figure 1. Left preauricular area edema



Figure 2. Leftside mandible deviation during mouth opening

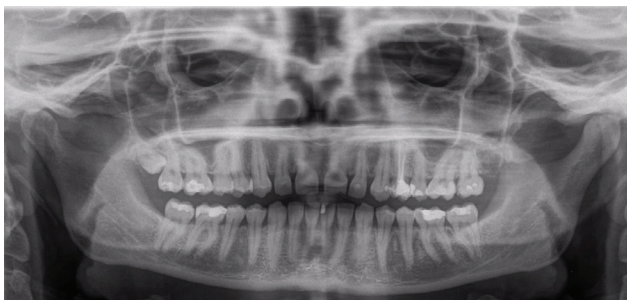


Figure 3. Preoperative OPG taken during clinical trial at Maxillofacial Surgery Outpatient Clinic

In order to broaden the diagnostic process with soft tissue imaging a MRI has been performed (Figure 4). T2-weighted images showed multiple, variform cartilaginous bodies inside the left temporomandibular joint. Also a large widening of articular capsule was present, affecting the profound masseter muscle layer and lateral pterygoid muscle (Figure 5).

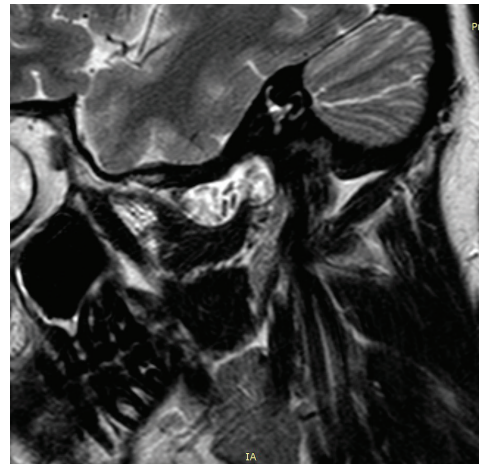


Figure 4. MRI - saggital scan through left TMJ, cartilaginous bodies visible in the articular cavity

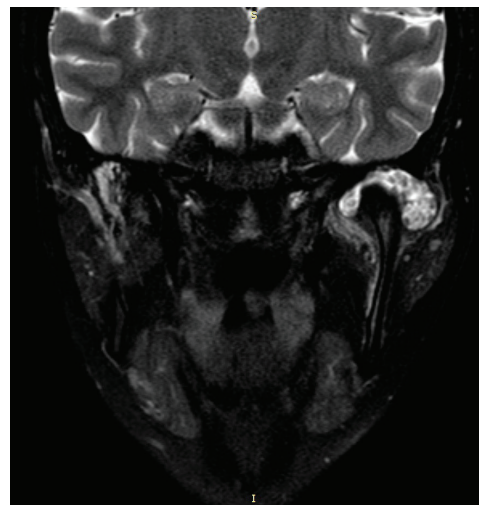


Figure 5. MRI - frontal scan through left TMJ, cartilaginous bodies visible in the articular cavity

Basing on physical examination and mentioned above imaging studies the patient was positively qualified for open joint surgery. The surgery has been performed under general anesthesia and facial nerve function monitoring and involved reaching to the articular capsule space via preauricular access. During the surgery the articular space has been irrigated and multiple variform, cartilage-resembling bodies were removed. They were not bounded to articular capsule and their biggest longitudinal dimension was about 3 mm (Figure 6). Right after the procedure the patient did not report significant relief which was because of post-treatment swelling.



Figure 6. Cartilaginous bodies removed from left TMJ during the first surgery



Figure 7. Postoperative control OPG.

The patient left the hospital in good general and local condition. During post-operative checkups the patient reported total regression of primarily presented symptoms. The obtained pathology report confirmed the initial diagnosis of synovial osteochondromatosis. About 4 months after the surgery the patient noted recurrence of pain and mandible lateral movement impairment. Physical examination confirmed left temporomandibular joint oedema reappearance, however a postoperative OPG did not reveal any abnormalities (Figure 7). A decision about subsequent hospitalization was made in order to perform next surgical procedure.

The surgery was again performed under general anesthesia and facial nerve function monitoring. It involved left TMJ synovial cavity revision and irrigation. Once more some cartilage-resembling bodies were removed. This time they were less numerous but resembled the ones received during the first surgery. The patient has been discharged from hospital again in well general and local condition. During the covid-19 pandemic the patient abandoned prescribed outpatient checkups.

Discussion

The necessity of second surgical intervention and lack of trauma in the past confirm primary form of synovial osteochondromatosis which is more aggressive and has a higher risk of recurrence than the secondary

form. Multiple research papers concerning hip joint synovial osteochondromatosis show that there is a 19% chance of recurrence in the same joint⁸. Long-term (7 years) disease evolution is characteristic for secondary form, however it must be noted that, according to Milgram, 3 phases of SOC development may be distinguished - initial, transitional and final. The third phase is the one when the loose cartilaginous bodies calcifies or ossifies therefore longtime presence of symptoms in this case was rather caused by diagnostic problems than etiology and form of the patient's disease^{4,6}. International researches suggest that proper diagnosis is stated the most often (over 80% of cases)⁵ in the third phase of SOC, however a thorough analysis of radiographic examination of patients reporting popular temporomandibular joint complaints (e.g. mouth opening difficulty or pain during chewing) may help in proper diagnosis and earlier treatment undertaking⁹.

Around 67% of patients suffering from synovial osteochondromatosis present mutation of gene *FNI* and/or *ACVR2A*, what is more - molecular tests showed that 56% of patients presents fusion of those 2 genes. Worth noticing is the fact that such a gene fusion is also characteristic for synovial chondrosarcoma which radiographic image may resemble synovial osteochondromatosis¹⁰. Apart from that, *ACVR2A* gene mutation (which is involved in skeletal system development) is also connected to large intestine carcinoma, hepatocellular carcinoma and cholangiocarcinoma¹¹.

Conclusions

Due to these reports about molecular background and possibility of occurrence of other diseases induced by the same gene mutation it seems to be crucial to perform further researches that would help to identify all the factors that may lead to synovial osteochondromatosis onset, especially the primary form. Also surgical treatment effectiveness assessment should be performed as it is the treatment of choice with no alternative in conservative treatment.

References

1. Zhao W, Ruan Y, Zhang W, Yang F. Synovial chondromatosis of the temporomandibular joint with 400 loose bodies: a case report and literature review. *J Int Med Res*, 2021;49:3000605211000526.
2. Grace MM, Letonoff EJ. Synovial Chondromatosis: An Unusual Case of Knee Pain and Swelling. *Fed Pract*, 2018;35:45-48.

3. Pinto AA Jr, Ferreira e Costa R, de Sousa SF, Chagas MR, do Carmo MA, de Lacerda JC. Synovial Chondromatosis of the Temporomandibular Joint Successfully Treated by Surgery. *Head Neck Pathol*, 2015;9:525-529.
4. Sozzi D, Bocchialini G, Novelli G, Valente MG, Moltrasio F, Bozzetti A. A rare case of synovial chondromatosis of the inferior TMJ compartment. Diagnosis and treatment aspect. *Ann Stomatol (Roma)*, 2016;6:91-95.
5. Nath P, Menon S. Synovial Chondromatosis of the Temporomandibular Joint. *J Maxillofac Oral Surg*, 2020;19:230-234.
6. Dwidmuthe S, Sharma M. A Case Report of Primary Synovial Chondromatosis with Bilateral Genu Valgum. *J Orthop Case Rep*, 2017;7:92-95.
7. Whyte A, Boeddinghaus R, Bartley A, Vijeyaendra R. Imaging of the temporomandibular joint. *Clin Radiol*, 2021;76:76.e21-76.e35.
8. Van der Valk MR, Veltman ES, Assink J, Veen MR. Synovial chondromatosis of the hip- a case report and literature review. *J Orthop*, 2019;16:249-253.
9. Barraclough O, Wilson G, Power A. Synovial chondromatosis of the temporomandibular joint: a case report. *Ann R Coll Surg Engl*, 2020;102:e213-e215.
10. Agaram NP, Zhang L, Dickson BC, wanson D, Sung YS, Panicek DM, et al. A molecular study of synovial chondromatosis. *Genes Chromosomes Cancer*, 2020;59:144-151.
11. Lasmar NP, Vieira RB, Rosa Jde O, Lasmar RC, Scarpa AC. Synovial chondromatosis. *Rev Bras Ortop*, 2015;45:490-492.

Received on April 25, 2023.

Revised on May 20, 2023.

Accepted on May 26, 2023.

Conflict of Interests: Nothing to declare.

Financial disclosure Statement: Nothing to declare.

Human Rights Statement: All the procedures on humans were conducted in accordance with Helsinki Declaration of 1975, as revised 2000. Consent was obtained from the patient/s and approved for current study by national ethical committee.

Animal Rights Statement: None required.

Correspondence:

Jakub Wójcicki

Maxillofacial Surgery Ward of 10th Military Research Hospital and Polyclinic

Bydgoszcz, Poland

e-mail: chirurgia_szczekowa@10wsk.mil.pl