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CASE REPORT

A case report of a primary synovial chondromatosis of temporomandibular joint, producing medial displacement of the articular disc

C. Droguett[1,2,3], J.E. Fumeaux[3,4] & N.P. Skármeta[5,6]

[1]Universidad de Chile, Santiago, Chile
[2]Hospital del Trabajador, Santiago, Chile
[3]Department Oral and Maxillofacial Surgery, Universidad Mayor, Santiago, Chile
[4]Universidad Finis Terrae, Santiago, Chile
[5]Universidad Mayor, Santiago, Chile
[6]Department of Orofacial Pain, Universidad del Desarrollo, Santiago, Chile

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Correspondence to:
Dr. C. Droguett
Department Oral and Maxillofacial Surgery
Universidad Mayor
Santiago 7500857
Chile
Tel.: +562 27619309
Fax: +562 22403857
email: droguett.tidy@gmail.com

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Abstract

Synovial chondromatosis in the temporomandibular joint (TMJ) is a rare benign cartilaginous metaplasia characterized by the formation of loose bodies within the synovial joint space. This condition presents a specific triad of semiological characteristics featuring pain, swelling, and limitation of function. We will present a clinical case of a 49 years-old female patient who presented to the clinica Davila's maxillofacial service with severe functional alterations in her right TMJ. Clinically, the patient had a maximum aperture of 15 mm wide, alteration of mandibular dynamics and preauricular pain. Posterior conducting a complete clinical examination and conclusive magnetic resonance imaging (MRI) findings it was decided to perform surgery on the joint. Articular cavity was exposed through a preauriclar approach, where the joint capsule presented no evident alteration. After opening the capsule, we were able to identify and extract multiple cartilaginous bodies both above and below the disc space. The articular surfaces were in good condition, while the articular disc was shifted to the medial portion of the cavity. The removal of the loose bodies was held the meniscopexy of the articular disc with a small joint anchor. The immediate post-operative period was uneventful, and the patient did not present any alteration with respect to motor function of facial expression. At the 6-week control, we recorded an increase in maximum mouth opening of 30 mm and a reduction of the pain. Six months later, we recorded an even wider mouth opening with no signs or evidence of recurrence.

Introduction

Synovial chondromatosis (SC) is a very uncommon monoarticular non-neoplastic joint condition that usually affects large synovial joints. It is a cartilaginous metaplasia of the residuary mesenchyma in the synovial tissue, characterised by the formation nodules within the subsynovial connective tissue. These cartilaginous formations may be partitioned, calcified or form loose as free floating bodies in the intracapsular space.

Laennec first recognised SC in 1813. Since then, the cases revealed in literature usually described involvement of large synovial joints such as hip, elbow, knee, wrist, ankle and shoulder[1]. In 1933, Axhausen described the first case SC affecting the temporomandibular joint (TMJ)[2].

The etiology of SC of the TMJ is still unclear. Nevertheless, cases are commonly divided into primary and secondary SC. Primary cases cannot be associated with any identifiable etiological factor. Secondary SC cases are frequently associated with inflammatory joint disease, non-inflammatory arthropathy, repetitive microtrauma and previous trauma.

Guarda-Nardini *et al*. describe that this condition usually features a specific triad semiological characteristics such as unilateral pain, joint swelling and limitation of function[3]. Also, SC of the TMJ may present crepitation, limited opening and occlusal changes.

The clinical diagnosis of the SC should be made mainly based on imaging findings. Conventional imaging may show widening of the joint space and bony erosion, but are unable to detect the cartilaginous nodules. Computerised tomography and magnetic resonance imaging (MRI) have proven to be useful in diagnosing, surgical treatment planning and follow-up of SC[4].

The definitive diagnosis of SC of the TMJ relies on the histological analysis, and the treatment includes the removal of the loose bodies together with the affected synovium or disc.

The aim of this study is to present a case of an intracapsular primary SC of TMJ affecting the superior and inferior joint space producing medial displacement of the disc, without any further compromised structures.

Case report

A 49-year-old female patient was referred to Clinica Davila's maxillofacial service and presented with severe functional alterations in the right TMJ. Clinically, the patient had a maximum aperture of 15 mm with slightly right side-deflected mandibular dynamics. Also, she presented a slight swelling on the preauricular area and pain quantified by visual analogue scale (VAS) = 8.

Subsequent to a careful clinical examination and an MRI, it was decided to perform surgery on the affected joint. The articular cavity was exposed through a preauricular approach, in which it was observed that the joint capsule presented no evident alteration (Fig. 1). After opening the capsule, we were able to identify and remove multiple cartilaginous bodies both above and below the disc space(Figs 2–4). The articular surfaces were in good condition, while the articular disc was displaced to the medial portion of the cavity. After the removal of the bodies, a meniscopexy of the articular disc was performed with a small joint 5 mm anchor Mytek 2.0 mini (Mitek Products Inc., Westwood, MA, USA) and Etibon 2.0 (Ethicon, Inc., Somerville, NJ, USA). In the immediate post-operative period, the patient did not present any alteration concerning motor function or facial expression. In the next control (6 weeks post-surgery), the patient presented an increase in maximum jaw opening of 30 mm and the pain scores were significantly lower (VAS = 0–3).

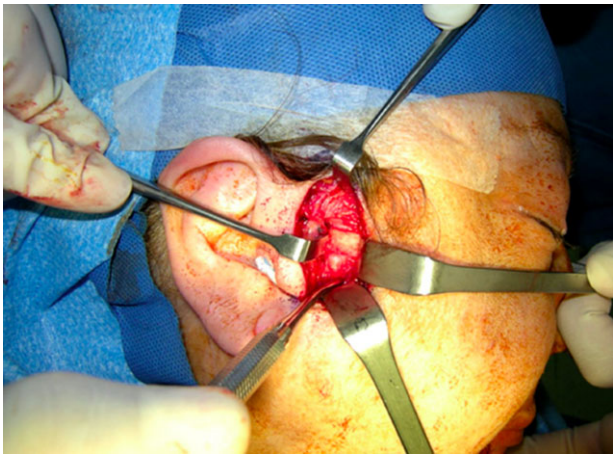

Figure 1 Exposure of the joint capsule through a preauricular approach.

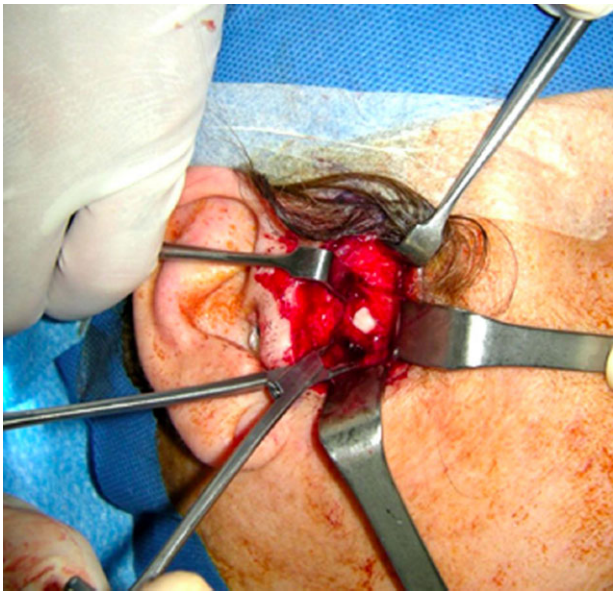

Figure 2 Exposure of a calcified loose body through the incision on the joint capsule.

Follow-up

The post-operative was uneventful, and the following assessment was scheduled at 6 weeks, 6 months and

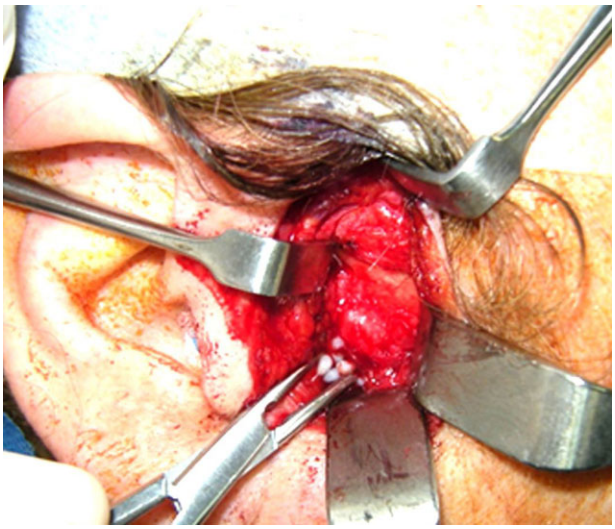

Figure 3 Removal of cartilaginous calcified loose bodies.

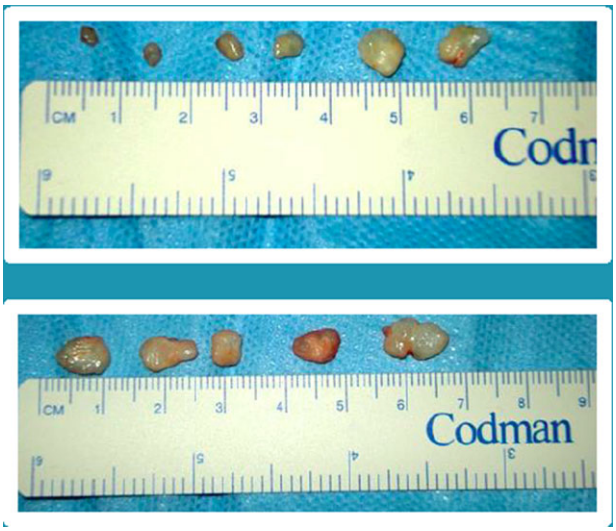

Figure 4 This SC case was characterised by the presence of multiple loose cartilaginous bodies. SC, synovial chondromatosis.

1 year post-surgery. At the 6-month appointment, a maximum mouth opening of 41 mm was recorded, without any signs of recurrence.

Histopathologic analysis

The macroscopic examination showed irregular nodules of a grayish-brown tissue of 0.3–0.8 cm. The following microscopic analysis showed nodular fragments of cartilaginous tissue with stromal hyaline degeneration and the presence of dystrophic calcifications.

Discussion

There are over 200 case reports of SC affecting TMJ described in literature[5]. Most of these descriptions were confined to the superior articular compartment, affecting the superior synovial lining.

It has been suggested that one of the main reasons for this characteristic presentation is the distinctive anatomic dimensions between the superior and inferior compartment[6]. The inferior compartment being much narrower than the superior compartment consequently will have a less extensive area of synovial membrane. Holmlund *et al*. weighted that loose bodies in the inferior articular space might have originated on the superior synovial lining and settled down on the lower articular compartment following the perforation of the disc[7]. In this regard, we believe that it may be possible that the settling of the cartilaginous bodies within the superior subsynovial space might have had some inherence on compromising the lateral collateral ligament, shifting the articular disc towards an anterior and medial position and enabling the loose bodies to occupy the inferior articular space.

The unspecific symptomatic manifestations produced by SC are usually hard to relate to a specific diagnostic. Alteration of the masticatory function, pain, swelling, crepitation and temporary disruption of the occlusion often mislead the clinician. These disturbances are commonly uncovered in degenerative inflammatory processes of the TMJ such as osteoarthritis, making misdiagnosis or late diagnosis very frequent[8]. Nevertheless, modern MRI techniques make a significant diagnostic contribution in the diagnosis of SC, revealing structural changes in the TMJ capsule, synovium, disc and medullary cavities and showing inflammatory reactions such as effusion. Also, it allows identifying hypodense semi-calcified loose bodies and their relation with subjacent structures[9]. The MRI findings on this particular case show a synovial proliferation with punctuate calcifications on the right TMJ using a T2-weighted coronal sequence (Figs 5).

Due to SC is a chronic and progressive metaplastic condition, that does not seems to undergo on a spontaneous resolution. Removal of the loose bodies is often the most selected and recommended therapeutic choice. Consequently, the most common approach chosen by authors is open access surgery featuring partial synovectomy. Other procedures in addition to the removal of the cartilaginous proliferation were also described, such as total synovectomy, discectomy and condylectomy, but they were less frequently indicated[3,10,11].

Figure 5 MRI revealing the synovial proliferation in the right TMJ with punctate calcifications, displacing the articular disc to a medial position. MRI, magnetic resonance imaging; TMJ, temporomandibular joint.

More conservative techniques are also mentioned in literature, such as two-needle arthrocentesis or minor arthroscopic surgery[12,13]. Nevertheless, these less aggressive procedures have low success rates and usually concluded in open access surgery[3]. Honda *et al*. suggested that an arthroscopic approach should be considered when the loose bodies have a size less than 3 mm wide[12]; however, current data support that open access surgery remains the most certain therapeutic approach. During the intraoperative procedure and after removing the loose bodies, the TMJ structures were carefully inspected, not finding any signs of inflammatory disturbances on the synovial membrane and not finding evidence of erosion on the TMJ surfaces or abnormalities on the articular disc.

The definitive diagnosis can be made through arthroscopy or during the intraoperative procedure, but it must be confirmed through the histopathological analysis. Miligram on a histopathologic study manage to categorise SC in three different stages. The first stage is characterised by metaplastic activity of the synovial tissue without formation of loose bodies. During the second stage, there is presence of loose bodies plus metaplastic nodules of the synovial tissue in a proliferative state, and in the third stage there are multiple loose bodies with no metaplastic activity signs or synovium alterations[14]. It has been suggested that in more advanced stages of this disease, in which calcification of the cartilaginous bodies has taken place and the synovial lining remains inactive, less aggressive surgical procedures should be considered because of the possibility of the synovial membrane not being in proliferative state[15]. Also, it has been documented that calcification or ossification of the cartilaginous formations usually damages the articular disc and/or can compromise extra-articular structures[16].

In this particular case, no extra-articular structures were compromised. After completed intraoperative inspection, it was advocated to maintain the structural functionality of the TMJ; therefore, we decided to conserve the synovium and performed the meniscopexy of the medially displaced disc.

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