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Synovial Chondromatosis of the Temporomandibular Joint with Extension to the Glenoid Fossa

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Abstract **Case Report**

Synovial chondromatosis is a rare pathology that affects the large joints, it is exceptional in TMJ, it is related to a synovial metaplasia with production of cartilaginous bodies in the temporomandibular joint **(**TMJ) , its slow evolution and its non-specific clinical presentation are the causes of the diagnostic delay that characterizes this pathology; We present a case of synovial chondromatosis of the TMJ in a 38 year old patient, in whom the CT scan showed a cartilaginous tumor of the TMJ with erosion of the glenoid fossa without intracranial extension, The biopsy with anatomopathological study confirmed the diagnosis of synovial chondromatosis, the loose cartilaginous bodies were completely cleaned by arthrotomy through a right preauricular approach, the evolution was good, and the two-year follow-up did not find any sign of recurrence. Imaging, especially MRI, is very useful in the diagnosis of this pathology; there is no consensus regarding the treatment of synovial chondromatosis, either by arthrotomy or arthroscopy, the main point is to clean all loose cartilage bodies to avoid recurrence.

Keywords: Synovial chondromatosis, MRI, anatompathological study, arthrotomy, arthroscopy.

INTRODUCTION

Synovial chondromatosis (SC) is a rare articular pathology that affects the large joints (knee, hip, shoulder...), and more rarely the temporomandibular joint (TMJ). It was described for the first time on the knee by Ambroise Paré in 1558, and in 1933 by Axhausen on the temporomandibular joint (TMJ); since TMJ tumors are basically unfrequent, SC is considered the most frequent benign tumor of the TMJ [1, 2].

This pathology is characterized by synovial metaplasia associated with the production of loose cartilaginous or osteocartilaginous bodies inside the joint cavity; two forms of Synovial chondromatosis have been described, primary and secondary, the pathogenesis of the primary form is unknown; hypotheses have been proposed to explain the secondary form: chronic inflammation, repetitive microtrauma, parafunctional habits of the temporomandibular joint [3- 5].

Considering its slow evolution associated with a nonspecific clinical manifestation, synovial chondromatosis is mostly diagnosed at delayed stages. radiological investigations: orthopantomogramm, CT scan and MRI allow guiding the diagnosis; the latter is only confirmed by histopathological study.

Treatment is essentially surgical by arthrotomy and/or arthroscopy; recurrences are rare but may occur,and malignant transformation into chondrosarcoma is possible but remains exceptional [6, 7]

We report a case of a synovial chondromatosis of the right TMJ, with extension to the cranium base, which was managed by arthrotomy, with good clinical evolution, without recurrence after 24 months follow-up.

OBSERVATION

A 38 year old female patient, without any particular pathological history, especially of the temporomandibular joint, attended the maxillofacial and oral surgery consultation of the Mohamed VI University Hospital of Marrakech; The main complaint was pain and swelling of the right preauricular region (Figure 1). The swelling was firm and fixed to the deep plane, without any skin anomalies, evolving very

slowly over the last 4 years, associated with a latero-deviation of the mandible towards the left side at mouth opening; there was no crepitus, no dislocations and subluxations, no limitation of mouth opening, and no intracranial hypertension signs.

The CT scan of the facial massif showed the presence of a peripheral multi-lobulated intra-articular tumor, causing an important widening of the inter-articular space, this process surrounds - from all sides - the mandibular condyle and the upper part of the Ramus (Figure 2), a thinning of the homolateral temporal eminence was observed with an endocranial bulge with no infiltration of the cerebral parenchyma (Figure 3).

CT scan suspected a cartilaginous or synovial origin of the tumor, which motivated a surgical exploration and biopsy through a preauricular incision, this exploration found a multi-lobulated whitish process, whose aspect reminds the aspect of cartilage, these lobules were easily detachable with the curette. The anatomopathological study showed an aspect in conformity with a synovial chondromatosis of the right TMJ.

Arthrotomy was decided, the temporomandibular joint was approached via the classic pre-auricular approach, the elevation of the musculocutaneous flap anteriorly made it possible to visualize the mass, the joint capsule could not be located, the mass was whitish, made of an aggregation of multiple easily detachable rounded bodies, whose appearance was cartilage-like (Figure 4); with the help of an elevator, the mass was detached from the adjacent structures, the synovium and the articular disc were incorporated in the mass, the mandibular fossa was largely eroded, without communication with the endocranial space, its surface was delicately scraped with a curette to remove all loose bodies; access to the anteromedial face of the condyle was possible, facilitated by the space created by the erosion of the mandibular fossa and the removal of the disc and synovium; the condyle was judged to be intact and left in place; the partially eroded mandibular fossa was sufficiently solid (Figure 5), therefore no reinforcement was performed.

The postoperative recovery was smooth, the pain was effectively managed by level 1 and 2 analgesics; the patient was advised to have a mixed diet the first weeks after surgery.

The follow-up at 24 months, noted a symmetric face without swelling or pain, with normal mouth opening and good occlusion (Figure 6 and 7).

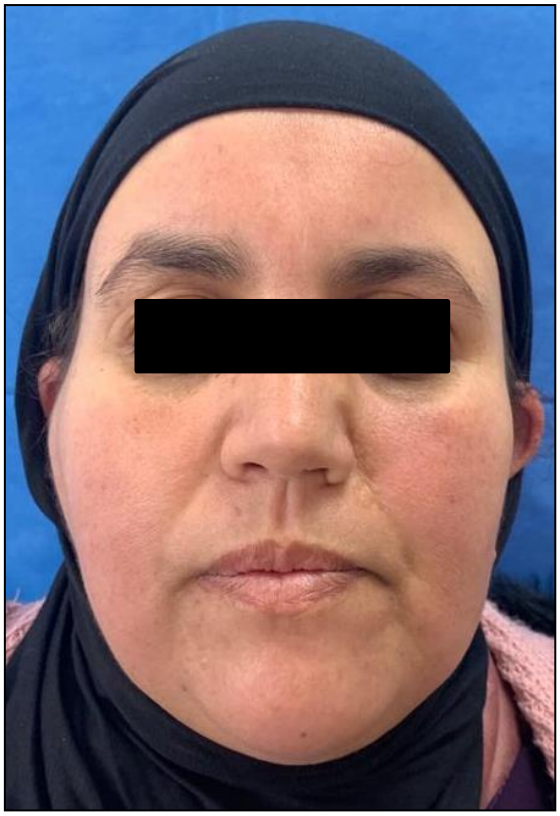

Figure 1: Image of the patient showing the swelling in the right preauricular region

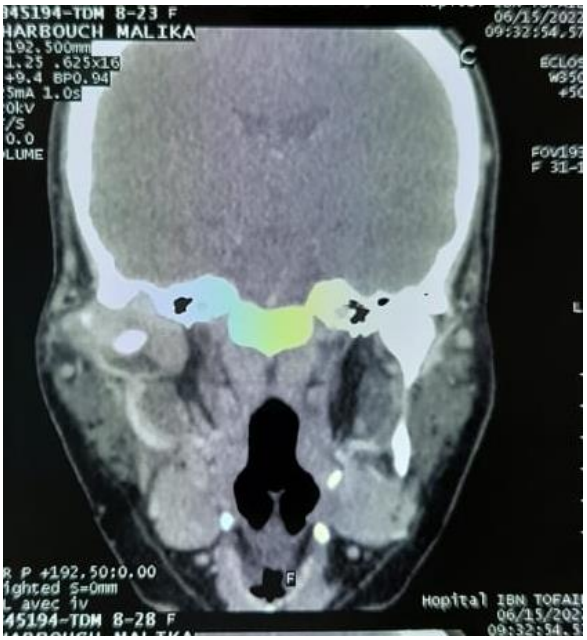

Figure 2: CT scann showing intra-articular tumor, causing an important widening of the inter-articular space of the right TMJ

Figure 3: Thinning of the homolateral temporal eminence without infiltration of the cerebral parenchyma

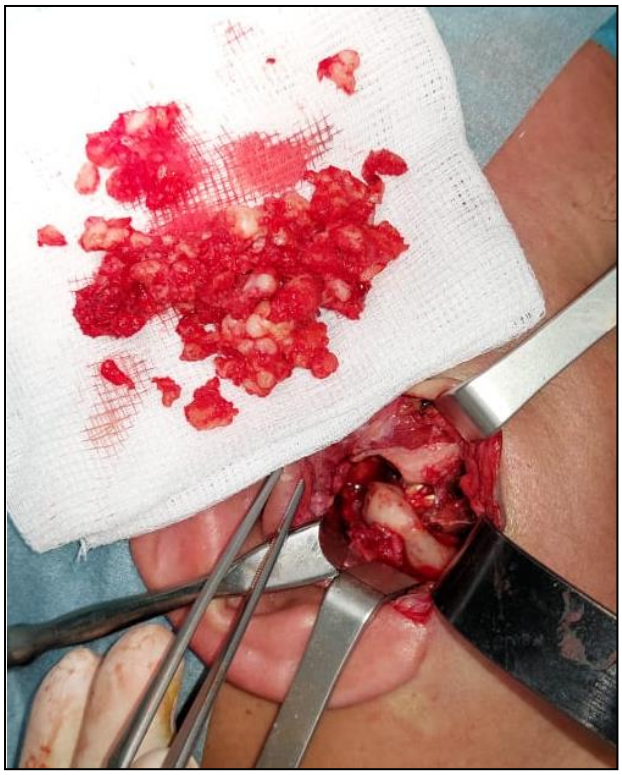

Figure 4: Intraoperatively image showing: multiples whitish easily detachable rounded bodies, whose appearance was cartilage-like

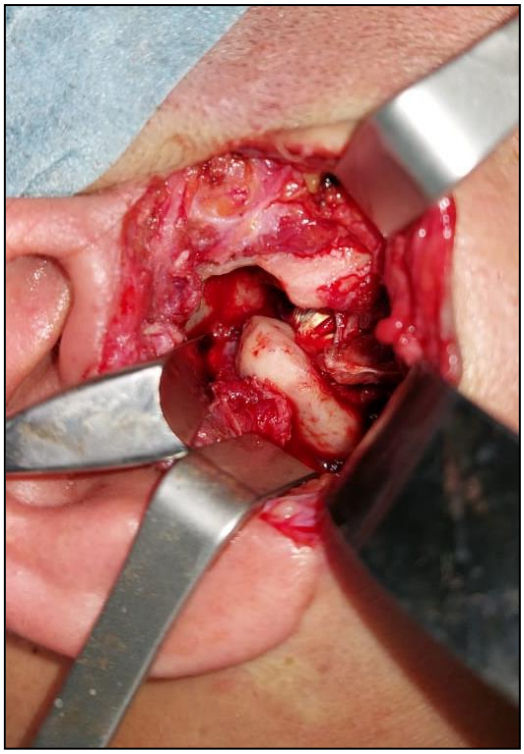

Figure 5: The appearance of the TMJ joint after cleaning of all loose bodies, the condyle looks intact, the glenoid fossa looks eroded

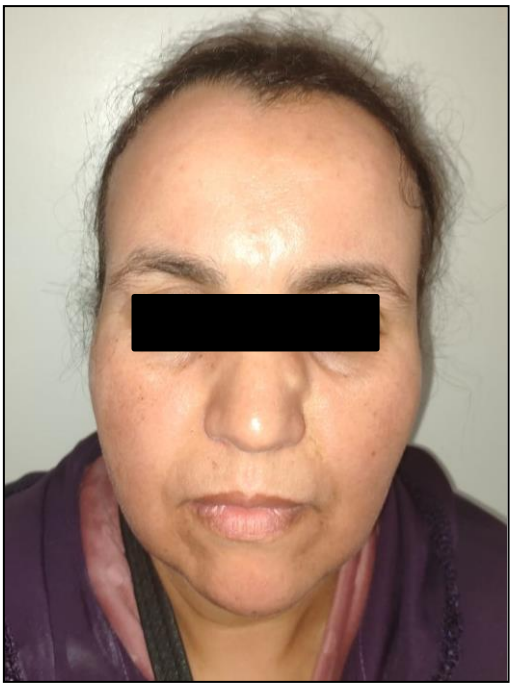

Figure 6: Photo of the patient 2 years postoperatively, showing a symmetric face, without swelling

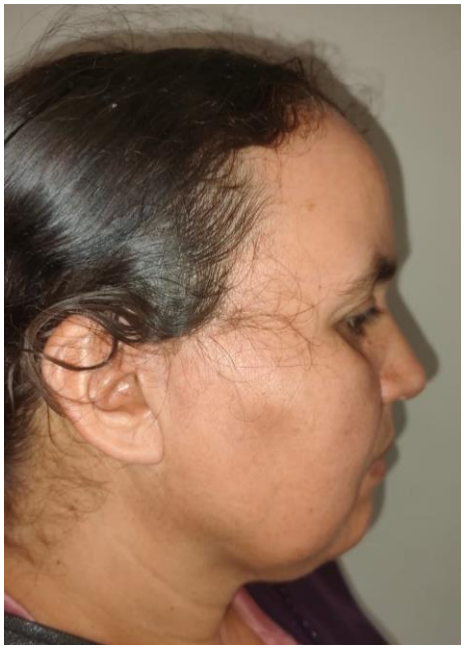

Figure 7: Right profile photo: showing the absence of swelling and the absence of scar

DISCUSSION

Synovial chondromatosis, also called osteochondromatosis or chondrometaplasia, is a rare benign pathology of the large joints, and even rarer in the temporomandibular joint, however, there has been a marked increase in the number of published cases over the past decade, it appears that CT and MRI have played an important role in the increase in diagnosed cases [6, 8, 9].

TMJ SC affects mostly young women [6], is characterized by an abnormal proliferation of the synovium, associated with the formation of cartilaginous bodies or chondromes, which pediculate and detaches to become lodged in the joint, and may later calcify to form ostéochondromes [10].

The clinical presentation is dominated by nonspecific signs, such as pain, preauricular swelling, mouth opening limitation, malocclusion and crepitus. This nonspecific clinical expression and the slow evolution, lead to the delay in diagnosis that characterizes this pathology [6, 11, 12]; The majority of patients are diagnosed at stage 3, according to the classification of Milgram [10, 13]; considering the poor value of the clinical presentation of synovial chondromatosis, radiological investigations are necessary and helpful to the diagnosis, especially MRI and CT scan, which do not always show specific signs of the pathology; the CT can visualize the cartilage bodies, but it is more performant in studying bonny structures, like the condyle and glenoid fossa [8].

MRI is the reference imaging examination [6], it allows visualization and localization of osteochondromas in the joint, and also allows to show indirect signs of joint suffering such as joint effusion [2], and allows to study the extension of the lesion towards the surrounding structures (base of the cranium, infra temporal fossa, pterygoid muscle, temporal eminence, parotid...) [2].

The radiological findings of SC can be very evocative; however, several authors recommend a biopsy with a histological study to confirm the diagnosis before deciding on the definitive treatment. The aggressiveness and local lysis that characterize SC sometimes suggest a malignant pathology, the management of which is totally different [10, 14, 15].

In our case and considering the aggressive radiological aspect of the process, we opted for a surgical biopsy before deciding the treatment option.

Milgram [13] was the first to establish a staging of this pathology, based on histological study, thus he described:

Stage 1: Initial stage: intra-synovial metaplasia without loose bodies in the joint;

Stage 2: Transitional stage: active synovial chondrogenisis and cartilage nodules or loose bodies are released into the articular cavity.

Stage 3: Chondrogenisis is stoped, multiple chondromas are seen in the joint, which may calcify secondarily.

Synovial chondromatosis is a pathology that does not resolve spontaneously nor under medical treatment; therefore its management is essentially surgical [12, 13, 16-18]. The treatment consists of the removal of all loose bodies from the inside of the joint, associated or not with synovectomy and/or resection of the articular disc, or even with condylectomy.

Surgical techniques are divided into invasive procedures, which is essentially arthrotomy, and less invasive procedures, represented by arthroscopy and arthrocentesis; the choice of the technique to be used is controlled by several elements: the stage of the disease according to the Milgram classification [19, 20], the location, size and abundance of loose bodies, and also the extension to adjacent structures [7].

Arthrotomy or open surgery is the most invasive method, yet it remains the option of choice for SC stage 1 or 2, requiring in addition a synovectomy [3], or with loose bodies of significant abundance and size (>3mm) [12] or in the both compartments of the temporomandibular joint [7], the extension of the process, particularly to the base of the cranium and in the endocranial region, cannot be controlled endoscopically [21, 22], especially if temporal fossa reconstruction, temporal muscle transposition or condylectomy are planned [7, 23, 8, 12]. Access to loose bodies in the anteromedial region of the temporomandibular joint may be difficult through

arthrotomy, which has motivated some authors to propose condylectomy or amputation of the zygomatic arch for complete cleaning [6, 24].

Arthroscopy appears to be less chronophonic and less invasive [25] and therefore carries less risk mainly for the facial nerve [12, 26]. And finds its place essentially in case of SC stage 3, with loose bodies of small size an number, lodged in the upper compartment of the temporomandibular joint, arthroscopy also allows according to series the electrocoagulation of the zones of synovial metaplasia as well as the articular ligaments [7, 16, 21]. Although, the removal of loose bodies of great abundance or considerable size >3mm [11, 27], or synovectomy remain difficult or even impossible to perform endoscopically, and sometimes require the use of larger cannulas, additional incisions or splitting of the giant bodies, or even have recourse to arthrotomy [6, 7, 21].

In our case, arthrotomy was chosen, considering the large size of the tumor, its location in both compartments of the temporomandibular joint, the extension to the temporal bone, and the experience of the surgeon.

The combined technique (open surgery and arthroscopy) has also been described. This technique allows the removal of all chondromas without the need for large osteotomies and in a shorter operating time [17, 28].

The evolution is generally favorable, recurrence may be correlated to an incomplete resection or to active metaplasia inside the bone erosions [6], and can lead to more aggressive surgery; fortunately risk of recurrence is very low [7, 11, 14, 21]. Malignant transformation is exceptional [14, 29, 30]. Long term follow-up is recommended [3].

CONCLUSION

Synovial chondromatosis in TMJ is a rare and long termed silent disease, its clinical presentation is not pathognomic, therefore, it is usually unnoticed, misdiagnosed or diagnosed with delay; MRI and CT scan are very helpful to guide the diagnosis; confirmation is obtained by histological examination, of the specimen; SC treatment is surgical removal of all loose bodies, by arthrotomy or arthroscopy.

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