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Case Report

Synovial Chondromatosis of the Temporomandibular Joint – Clinical, CT, MRI, Surgical and Histological Findings: Case Report

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ABSTRACT

Synovial chondromatosis is a benign metaplastic disorder, rarely located in the temporomandibular joint. The present study describes a 50-year-old woman with unilateral pre-auricular pain, crepitus and swelling of the area. The clinical, imaging, surgical and histopathological findings established the diagnosis of unilateral TMJ synovial chondromatosis.

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Introduction

Synovial chondromatosis (SC) is a benign metaplastic disorder of unknown etiology that affects any synovial joint, but only 3% of the cases include the temporomandibular joint (TMJ) [1]. SC is characterized by the presence of multiple small foreign bodies within the joint spaces, chondral type bodies that compress and distended the joint capsule [2-4]. SC is originated from a proliferative disease of the synovial membrane. The nodules of the joint space remain in contact with the synovial membrane, and depending on the degree of evolution, SC interferes with free mandibular movement, gradually increases the volume of the region, tenderness, or intermittent joint pain, crepitus, and

deviation toward the affected side [5]. SC is usually monoarticular and most commonly affects large joints, but it also has been reported in the middle ear and the malleolar canal [6-8]. Initially, SC is characterized by a primary form, in which fragments of the cartilage beneath the synovial membrane undergo metaplasia and deposit chondromucin. These cartilaginous bodies enlarge and finally detach from the synovial membrane. The secondary form of SC occurs subsequent to pre-existing joint disease, such as arthritis or traumatism and involves synovial nourishment of dislodged soft tissue fragments [2].

The etiology of SC has been related to the possible role of traumatic events [9]. Clinical and pathological findings obtained from imaging

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studies play an important role in the early diagnosis of SC. TMJ most often affects people between the fourth and fifth decades of age and is more relevant in females (1.6:1 ratio). The most frequent location of TMJ is unilateral; bilateral involvement is rare and has also been reported in females, preferably located in the upper joint space [5, 10].

Conventional radiology and ultrasound have a limited role for SC diagnosis because they fail in the detection of intra-articular loose bodies in a significant number of cases, and additionally, not all calcifications are radiographically visible at early stages [11]. The present study describes a TMJ synovial chondromatosis and conducts a review of the literature on this disease.

Case Report

This report is part of the research project approved by the Ethical and Scientific Committee of the Universidad de Los Andes (CEC 201951), and informed consent was obtained from the patient.

A 50-year-old woman, with hypertensive blood pressure, well-controlled with nifedipine (Adalat®), an oral calcium channel blocker (10 ml/gr, three times a day) and acetylsalicylic acid (Aspirin®) as antiplatelet agent (100 ml/gr, once a day). Additionally, she presented mitral stenosis, and 10 years earlier, she used a trans-dermal gel (Ginoderm®) as estradiol hormone replacement therapy. She was originally derived for the study of the left parotid gland, not related to specific initial signs and symptoms that may mimic those of parotid gland tumors. The illness begun 5 years before, and she related it with trauma (family violence) in her left hemi-facial area. Clinically, she presented with unilateral swelling, pain, headache and noise during palpation; crepitus in the left TMJ appeared one year before. Her maximum forced open mouth was 40 mm, and a deformity in the left TMJ area was visually noticed. There were no neck adenopathies. Clinically could be confused with joint dysfunction, intracapsular bone fracture, avascular necrosis or arthritis, neoplasia or pre-auricular parotid mass.

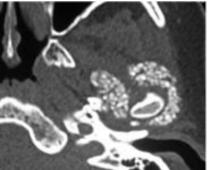


Figure 1: CT scan axial view showing multiple collections of little nodules surrounding the left TMJ, without deformation or remodeling of the condyle or tympanic plate.

CT exam was requested, identifying an expansion of the left TMJ, without the involvement of the bone or intracranial mass and accumulation of multiples radiopaque nodules inside the left TMJ, occupying all joint spaces around the condyle, including the upper region of the lateral pterygoid muscle, close to the pterygoid wing (Figure 1). The MRI examination confirms the findings, and these nodules caused capsule expansion in all directions (Figure 2).

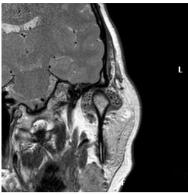


Figure 2: Coronal MRI view shows increased facial contour at the left parotid area, presenting joint spaces fully occupied by accumulation of multiple and little hypo intense nodules of franc hypo-signal, clearly capsulated, causing severe expansion of the joint capsule, structural integrity of the mandibular condyle and skull base, and the SC displaced the masticatory fat pack.

Before surgery, the cardiologist discontinued aspirin intake. The patient was operated under general anaesthesia with nasotracheal intubation; the approach was made in the pre-auricular zone, exposing the joint capsule. The capsule was opened, and it showed multiple white nodules; at this moment, the nodules projected outwards (Figure 3). Nodules showed a pearly appearance of various sizes and contour, hard consistency, smooth surface, and some of them and other hyalines were absolutely calcified; most of them were spherical and some oval shapes, accounting for approximately 320 units surrounding the condyle. Capsule thickness included multiple little nodules. The disc was observed thin and anteriorly positioned. The bony border of the TMJ was soft and smooth. Nodules were removed, underwent careful surgery cleaning, and abundant saline solution was used to rinse joint spaces; additionally, the abnormal synovial tissue, capsule and disc of the joint were fully removed, and suture was carried out by planes. Nodules, synovial tissue, and the capsule were sent for histopathological analysis (in 10% formalin solution, neutral buffered, Merck KGaA, Darmstadt, Germany).



Figure 3: Intraoperative open arthrotomy of the typical aspect of expanded capsule, and multiple nodules.

For post-surgical inflammation, prednisone was indicated for four days (in decreasing form, 20-15-10-5 mg per day) and Lysine Clonixinate (125 mg every 6 hrs) for 3 days. Additionally, the patient was indicated to intake Amoxicillin (1 g, four times a day, for 7 days) and Metronidazole (500 ml/g every 8 hrs, for 7 days). Ice wrapped in a towel was applied locally during the first 2 days, lateral to the surgical area.

In order to reduce the joint ankylosis risk, physiotherapy was carried out at 24 hours, including the use of chewing gum (Sugarless Orbit Gum®,

Wrigley's, Chicago, IL, USA) during 15 minutes, twice a day, after lunch and dinner. The patient was also instructed with guided exercises of open and closed mouth with hand resistance in the chin and included drawing a vertical line over the tip of the nose and chin to follow this axis during open and closed mouth exercises in front of a mirror. Another exercise was left and right movement with contra-lateral resistance using the ipsilateral hand (10 times each, 3 times a day, for 1 month). The first meals were ice cream and soft foods. The patient remained in the hospital for 3 days, including the entrance day. The suture was removed two weeks after the surgery, and evolution was without complications. She was controlled weekly for one month, and at present, she is controlled each year.

Microscopically, biopsy reported gross findings of connective tissue with multiple isolated nodules of chondroid matrix and calcified areas. The synovial cover was hyperplastic, with presence of enlarged chondrocytes, without atypical cells (Figures 4A-4D). The histopathological features provided the diagnosis of SC of the TMJ. Free-floating nodules were covered by a layer of the synovium. The subsynovial nodules of hyaline cartilage detached from the synovium to lie down within the spread joint space, and because cartilage is nourished by synovial fluid, the chondral bodies may grow and gradually increase in size in the joint space. Although the number of chondral bodies varies widely, they are most commonly numerous [12].

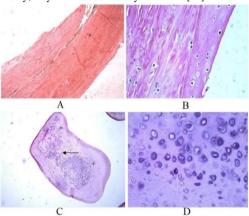


Figure 4: Histological observations of a gross connective tissue corresponding to synovial membrane with dense collagen fibers (Van Gieson Stain 4x) (4A). Chondroid differentiation of the synovial lining, (H-E Stain. 10x) (4B). A free nodule of hyaline cartilage with calcification areas (arrow), (H-E stain). 10x (4C). Nodule with typical chondrocytes in a chondroid matrix. Cells were regular in shape as seen with H-E stain, 40x (4D).

In the subsequent monitoring for 13 months, were observed absence of pain and crepitus, unforced achieved maximum mouth aperture of 58 mm, and joint noise disappeared completely after surgery. No recurrence was observed after clinical and CT control. Postoperative CT scan showed calcified nodules displaced to the pterygomandibular space, over the lateral pterygoid muscle. This was without clinical significance because it is not associated with synovial tissue and represents only the surgical difficulty to reach and remove nodules from this area (Figure 5).



Figure 5: Post-operative CT, one month after the surgery, showing a loose body at the left pterygomandibular space, without clinical manifestations and, apparently, without synovial tissue, but with thickening of the left temporal bone, compared to the contralateral side.

Discussion

TMJ SC was first described by Axhausen (1933) as a benign tumor [13]. However, the pathogenesis of SC still remains unknown, and it has been correlated with trauma, infection, embryologic disturbance, and parafunction as potential risk factors. Hopyan et al., based on the presence of recognized molecular abnormalities, believes that SC is a benign neoplasm, rather than a metaplastic illness [13-16]. Genetic features observed in the Hedgehog signaling pathway indicate that its target genes PTC1 and GLI1, which are typically involved in the development of other cartilaginous neoplasms, might play a role in SC development [16]. In addition, growth factors, such as fibroblast growth factor-2 and -3, change the expression in SC [17]. The proto-oncogene C-ERBB2 was found in a family case of two brothers with identical ankle SC. Finally, chromosome 6 abnormalities, identified at cytogenetic and molecular cytogenetic studies, have been a recurrent finding in SC; results could indicate that the disease is a neoplastic condition [18]. In the early stages, biomarkers related to mesenchymal stem cells, and chondrogenesis have been observed in SC [19].

In other joints, the literature describes the coalescence of multiple chondral bodies, creating a "giant" or "massive" firm appearance [20]. According to Milgram microscopic analysis, SC is composed of lobules of hyaline cartilage, surrounded by synovial lining (a two-cell layer of cuboidal epithelium). These pathologic features reinforce the use of the term SC, as emphasized by the World Health Organization nomenclature for this entity because the primary abnormality is sub-synovial cartilage metaplasia [21]. A variable degree of synovial proliferation or hyperplasia may also be present (Classified as Milgram stage 3).

Clinically, due to the gradual and chronic development, SC can be misdiagnosed as a lesion of the parotid gland, tumor of the preauricular region, dysplasia of the mandible, internal derangement, and osteochondromatosis [12, 22-24].

CT showed low attenuation of non-mineralized nodules of SC due to the associated joint fluid and the high-water content of the hyaline cartilage neoplastic tissues. CT is particularly helpful for finding characteristics of calcified nodule multiplicity [25]. In our case, the use of contrast media agents (CMA) in the CT scan was not justified because SC does not develop new blood vessels around the lesion, and the first image

series clearly showed typical nodules located in the expanded joint space without involving other tissues. The use of intra-articular CMA was not justified based on the high CT space resolution, and scanning was the best tool to improve SC identification, by adjusting the window level in order to obtain a bone window, enough to recognize high-density tissues of the area, and nodules not yet calcified. MRI was also performed in this case, clearly indicated the disease, recognizing capsules and synovial expansion, because those tissues were affected with enormous distension in all space directions. MRI, confirming the typical nodules reported as pathognomonic characteristics [22, 26]. SC images include chondral and calcified loose bodies in the TMJ spaces, but it is possible to also observe them in other joint pathologies, such as chondrosarcoma or benign inflammatory illnesses such as osteoarthritis, rheumatoid arthritis and avascular necrosis [25, 27].

Literature indicates that the treatment of choice for SC is surgical resection, and recurrences have not been reported in TMJ. Synovectomy and removal of nodules are related to optimal treatment of intraarticular SC [28].

Arthroscopic procedure in the TMJ has been considered as effective in the diagnosis and removal of loose bodies of synovial chondromatosis, and it appears to offer benefits in controlling pain and functional improvements, with low range of mouth opening limitations and complications. The achievement of arthroscopy is also very operator-dependent, and iatrogenic damage to the articular cartilage might occur, which can considerably modify the outcome; however, recurrences can be treated effectively with additional surgical intervention [20-30].

According to the literature, bony extensions of SC are sporadic and mostly occur in the superior joint space, and condylar erosion is extremely rare [4, 31]. In those cases, a differential diagnosis might consider the possibility of chondrosarcoma, but in this TMJ report, although it was a large chondromatosis, the condyle and temporal bone did not present bone erosions, or other morphological commitment.

The accuracy of anamnesis, the complementary examinations, and the multidisciplinary teamwork were the most important factors to achieve the diagnosis, treatment, and adequate prognosis for this patient. In this case, the early establishment of post-surgical mandibular dynamics by a kinesiologist (at 24 hrs.), helped to maintain maximum mouth opening and prevented opening limitations. The patient also received psychological support at the same health center, aimed at avoiding new episodes of domestic violence, empowering the patient with training courses to improve their economic independence.

Histologically, synovial chondromatosis is very similar to synovial chondrosarcoma, presenting loss of the clustering growth model, matrix with myxoid changes and necrosis, or cellular atypia [32, 33]. To establish the final diagnosis of the disease, it is mandatory to histopathologically verify a transitional change from fibrous connective tissue to cartilaginous tissue or chondrometaplasia [34].

Conclusion

Clinical information is not enough to define SC. A multidisciplinary approach was used in this report to detect and treat TMJ SC. The signs and symptoms were pain, swelling, and crepitus, which were nonspecific

and similar to other joint diseases. Moreover, the location in the preauricular area could have been misdiagnosed as a parotid neoplasm or another pathology, especially before performing image studies. CT scan provided optimal imaging to detect and characterize SC and the intraarticular calcification. MRI revealed low to intermediate signal intensity with T1-weighted scans and innumerable heterogeneous signal bodies that did not invade the middle cranial fossa, continuous hyposignal on the fossa, structural integrity, and normal condyle mobility; all this information was useful for differential diagnosis. The final diagnosis confirmation was obtained by the histopathological studies.

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