



CASE REPORT:

Synovial Chondromatosis of TMJ with Glenoid Dehiscence: Case Report

Condromatosis sinovial de ATM con dehiscencia glenoidea: caso clínico

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ABSTRACT: Synovial chondromatosis is a rare benign entity characterized by cartilaginous metaplasia of the synovial membrane. Its location in the temporomandibular joint (TMJ) is uncommon and, in elderly patients, mandates ruling out synovial chondrosarcoma. A 71-year-old woman, with no relevant comorbidities, presented with two years of right preauricular pain, mandibular deviation, joint crepitus, and a maximum mouth opening of 30 mm. Computed tomography revealed multiple intra-articular hyperdense bodies involving both supradiscal and infradiscal compartments, with mild condylar erosive changes. An open preauricular approach was performed, identifying approximately 20 cartilaginous loose bodies and a dehiscence of the glenoid fossa roof toward the middle cranial fossa, prompting subtotal excision without formal synovectomy. Histopathological and immunohistochemical study (S-100+, IDH-1–, Ki-67 ≈1%) confirmed calcified synovial chondromatosis, Milgram stage III, ruling out chondrosarcoma. At eight-month follow-up, the patient remained asymptomatic, with a maximum mouth opening of 42 mm (30 mm preoperatively) and no signs of reactivation. Bicompartamental involvement and glenoid roof dehiscence are uncommon findings that conditioned the surgical approach. Immunohistochemistry was decisive for ruling out chondrosarcoma. Subtotal management, although not generalizable, was pertinent in this specific anatomical scenario. This case highlights the importance of rigorous histopathological and immunohistochemical assessment and of modulating surgical management in the presence of complex anatomical findings in advanced synovial chondromatosis of the TMJ.

KEYWORDS: Chondromatosis Synovial; Temporomandibular Joint; Joint Loose Bodies; Glenoid Cavity; Immunohistochemistry; Case Reports.

RESUMEN: La condromatosis sinovial es una entidad benigna infrecuente, caracterizada por la metaplasia cartilaginosa de la membrana sinovial. Su localización en la articulación temporomandibular (ATM) es poco habitual y, en pacientes de edad avanzada, obliga a descartar el condrosarcoma sinovial. Mujer de 71 años, sin comorbilidades, con dolor preauricular derecho de dos años de evolución, desviación mandibular, crepitación articular y apertura bucal máxima de 30 mm. La tomografía computarizada mostró múltiples cuerpos hiperdensos intraarticulares con afectación supradiscal e infradiscal y leves cambios erosivos condíleos. Se realizó abordaje preauricular abierto, identificándose aproximadamente 20 cuerpos cartilaginosos libres y dehiscencia del techo de la fosa glenoidea hacia la fosa craneal media, por lo que se optó por exéresis subtotal sin sinovectomía formal. El estudio histopatológico e inmunohistoquímico (S-100+, IDH-1–, Ki-67 ≈1%) confirmó condromatosis sinovial calcificada en estadio III de Milgram, descartando condrosarcoma. A los ocho meses la paciente se mantenía asintomática, con apertura bucal de 42 mm (preoperatoria de 30 mm) y sin signos de reactivación. La afectación bicompartimental y la dehiscencia glenoidea son hallazgos infrecuentes que condicionaron la conducta quirúrgica. La inmunohistoquímica fue determinante para descartar el condrosarcoma. La actitud subtotal, no generalizable, fue pertinente en este escenario. Este caso ilustra la importancia del estudio histopatológico e inmunohistoquímico riguroso y de la modulación de la conducta quirúrgica ante hallazgos anatómicos complejos en condromatosis sinovial avanzada de la ATM.

PALABRAS CLAVE: Condromatosis sinovial; Articulación temporomandibular; Cuerpos libres articulares; Cavidad glenoidea; Inmunohistoquímica; Informe de caso.

INTRODUCTION

Synovial chondromatosis (SC) is an uncommon disorder characterized by cartilaginous metaplasia of the synovial membrane, with progressive formation of cartilaginous nodules and, in advanced stages, calcified or ossified intra-articular loose bodies. The World Health Organization classification (Soft Tissue and Bone Tumours, 5th edition, 2020) (1) includes it among benign cartilaginous tumours, recognizing its potential, albeit infrequent, for local recurrence and, exceptionally, malignant transformation to secondary synovial chondrosarcoma (1, 2).

The temporomandibular joint (TMJ) constitutes an unusual location for this entity, which predominantly affects larger joints such as the knee, hip and elbow (3). In the TMJ, SC shows a greater incidence in women, typically between the fourth and sixth decades of life, with usually unilateral involvement of the upper joint compartment (3, 4).

Its etiopathogenesis remains under debate, with hypotheses linking it both to a reactive phenomenon secondary to repeated microtrauma or chronic mechanical overload, and to a low-aggressiveness neoplastic process (3).

Milgram's histopathological classification, in force since 1977 (5), divides SC into three stages: Stage I, with active synovial metaplasia without loose bodies; Stage II, with coexistence of synovial proliferation and loose bodies; and Stage III, in which the synovium has ceased its metaplastic activity and the predominant finding is calcified or ossified loose bodies (5). It is important to note that this classification is strictly histopathological, and is decisive for definitive diagnosis, therapeutic decisions, and prognosis.

From a clinical standpoint, TMJ SC manifests with nonspecific symptoms such as preauricular pain, limitation of mouth opening, joint sounds and, occasionally, swelling, which overlap with those of

non-tumoral temporomandibular disorders, hindering early diagnosis (3, 4). Imaging studies with computed tomography (CT) and, complementarily, magnetic resonance imaging, are essential (4). Definitive diagnosis, however, rests on histopathological examination, which also allows ruling out the main differential diagnosis: synovial chondrosarcoma, particularly relevant in elderly patients (1, 2).

The treatment of choice is surgical excision of the loose bodies, with or without associated synovectomy (4, 6). However, the systematic indication of synovectomy, the optimal extent of resection and the management of residual bodies in advanced stages remain a matter of debate, particularly when complex intraoperative findings such as bicompartamental involvement or dehiscence of the glenoid fossa roof toward the middle cranial fossa are present (7, 8).

The present work describes a case of calcified TMJ synovial chondromatosis, Milgram stage III, with bicompartamental involvement and glenoid fossa dehiscence, in a patient treated at a public hospital with limited diagnostic resources. We analyze the clinical, imaging, histopathological and intraoperative findings, as well as the short-term functional outcome, in relation to current literature.

CASE REPORT

A 71-year-old woman, with no relevant comorbidities (ASA I), was referred to the Department of Oral and Maxillofacial Surgery of Hospital de San Fernando for right preauricular pain of approximately two years of evolution, associated with intermittent crepitant joint sounds.

Physical examination revealed pain on palpation of the right preauricular region and joint crepitation reproducible during mandibular movements. Maximum mouth opening was 30 mm, with mandibular deviation toward the right side during opening. Quantitative measurements of lateralities

and protrusion were not recorded preoperatively in the clinical chart, which constitutes a partial limitation of the present communication.

Computed tomography (CT) of the temporomandibular joint was performed as the imaging study. Due to availability limitations inherent to the public hospital setting, magnetic resonance imaging could not complement the study. CT revealed multiple intra-articular hyperdense nodular images distributed in both the supradiscal and infradiscal compartments of the right TMJ, findings consistent with calcified synovial chondromatosis. Mild erosive changes were also identified on the condylar surface, without evidence of frank bone destruction (Figure 1).

Given the chronicity of symptoms and imaging findings, surgical treatment was indicated. The procedure was performed under orotracheal general anesthesia via a right preauricular approach. Once the capsule was exposed and both joint compartments opened, approximately 20 cartilaginous loose bodies of pearly-white appearance and firm consistency were identified, ranging in size between 5 and 15 mm, distributed in both the supradiscal and infradiscal spaces, an uncommon finding in this entity. During dissection, a discontinuity of the synovial membrane occurred. After excision of accessible loose bodies, marked thinning of the glenoid fossa roof was identified by direct visualization, with an area of dehiscence toward the middle cranial fossa, without evidence of cerebrospinal fluid leakage or frank dural exposure. In light of this finding, formal synovectomy was not completed, and dissection in the medial recess was limited to avoid potential dural injury. Profuse irrigation of the joint space with physiological saline solution was performed, along with careful hemostasis and layered closure. No postoperative drainage was placed (Figure 2).

Histopathological examination of the surgical specimens described multiple firm, whitish-

grayish fragments with irregular surfaces, measuring $4.5 \times 4 \times 0.9$ cm in aggregate. Microscopically, well-defined nodules of hyaline cartilaginous tissue were observed in a multinodular arrangement, with mature chondrocytes housed in lacunae and mild cytological atypia, without necrosis or atypical mitotic figures. Foci of dystrophic calcification with incipient endochondral ossification were identified, along with reactive fibrous synovial reaction with chronic degenerative changes and areas of giant cell reaction. Immunohistochemical study showed nuclear and cytoplasmic positivity for S-100 in cartilaginous cells, negativity for IDH-1 and CKAE1/AE3, and a proliferation index by Ki-67 of approximately 1%. The histomorphological and immunohistochemical findings were conclusive for calcified synovial chondromatosis, reasonably ruling out the differential diagnosis of synovial chondrosarcoma.

The postoperative course was uneventful, with satisfactory healing of the surgical wound and resolution of the preauricular swelling and discomfort in the early postoperative period.

The immediate postoperative computed tomography evidenced a significant reduction in the

number of intra-articular loose bodies compared with the preoperative study. Small residual cartilaginous bodies persisted in the medial recess, without radiological signs of articular progression or reactivation. The thinning of the glenoid fossa roof previously observed intraoperatively was also confirmed (Figure 3).

At eight-month follow-up, the patient remained asymptomatic. Maximum mouth opening reached 42 mm, within the physiological range, with resolution of the functional limitation documented preoperatively (30 mm). Pain assessment using the Visual Analogue Scale (VAS) registered a value of 2/10 only at maximum opening, without spontaneous pain or pain reproducible on preauricular palpation. Absence of joint crepitation was noted, a sign present preoperatively (Figure 4).

The patient continues clinical and radiological follow-up every six months, in order to monitor the stability of residual bodies and detect early any sign of reactivation, articular progression, or recurrence, as recommended by current literature given the rarely reported risk of local recurrence and malignant transformation in chronic lesions.

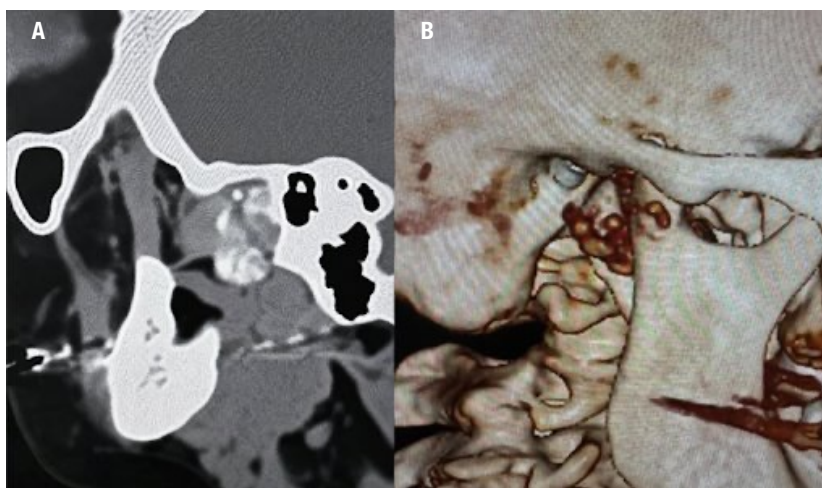


Figure 1. Preoperative imaging study. (A) Sagittal CT image of the right temporomandibular joint showing multiple hyperdense nodular structures (blue arrow) within the joint space, consistent with calcified cartilaginous loose bodies, involving both the supradiscal and infradiscal compartments. Mild erosive changes are observed on the condylar surface, without overt bone destruction. (B) Three-dimensional volumetric reconstruction showing the spatial distribution of the calcified intra-articular bodies and their relationship with adjacent bony structures.

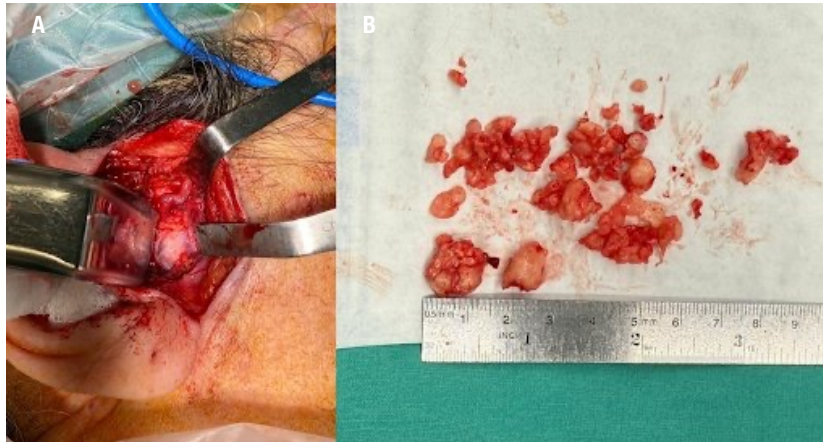


Figure 2. Intraoperative findings. (A) Right preauricular approach with surgical exposure of the joint: after layer-by-layer dissection and capsular opening, intra-articular loose bodies are visible. (B) Extracted cartilaginous loose bodies, whitish-grayish in color, with irregular surfaces and firm consistency, displayed on a sterile field next to a measurement ruler. Individual sizes range approximately between 5 and 15 mm; aggregated, the specimens sent for histopathological study measured 4.5×4×0.9 cm.



Figure 3. Immediate postoperative computed tomography. Coronal section at the level of the middle cranial fossa, showing marked thinning of the roof of the right glenoid fossa with an area of dehiscence (blue arrow) toward the middle cranial fossa. Small residual hyperdense bodies are also identified in the medial recess (green arrow), without signs of articular progression or reactivation.



Figure 4. Postoperative clinical outcome at eight months. (A) Right lateral view: healed preauricular scar without inflammatory signs or skin complications; preserved facial contour without asymmetry. (B) Maximum mouth opening of 42 mm, within the physiological range, without functional limitation or significant deviation; no audible joint crepitation on examination.

DISCUSSION

Synovial chondromatosis (SC) of the TMJ is an uncommon benign entity that, according to the 5th edition of the WHO classification of Soft Tissue and Bone Tumours (2020) (1), is included among benign cartilaginous tumours, with recognized risk of local recurrence and, in exceptional cases, malignant transformation to secondary synovial chondrosarcoma (1, 2). The patient in the present case has two demographic characteristics worth commenting on: female sex, consistent with the marked predilection described in the literature, with female-to-male ratios ranging from 1.5:1 to 4:1 in different series (3, 4), and an age of 71 years that exceeds the usual presentation range, located between the fourth and sixth decades of life (3, 4). This late presentation required a high index of suspicion for tumoral pathology during the diagnostic process, and reinforces the relevance of the exhaustive histopathological study performed.

A first noteworthy aspect is the value of histopathological examination for definitive diagnosis and Milgram staging (5). Although many published works classify TMJ SC cases by imaging alone, it should be remembered that Milgram's classification is strictly histopathological: it depends on the state of activity of the synovial membrane, not solely on the presence of calcified loose bodies (5). In the present case, the microscopic findings are consistent with Stage III. Available literature indicates that most TMJ SC cases are diagnosed at this advanced stage (4), attributable to the nonspecific symptomatology and slow clinical progression of the entity (3).

The obligatory differential diagnosis in a seventh-decade patient with cartilaginous synovial metaplasia is synovial chondrosarcoma, either primary or secondary to a long-standing SC (1, 2). Its exclusion cannot be based exclusively on morphological criteria, especially when mild cytological atypia is observed, as in this case.

Immunohistochemical study was decisive: nuclear and cytoplasmic expression of S-100 confirmed the cartilaginous origin of the lesion; negativity for IDH-1 argued against conventional chondrosarcoma, in which IDH1/IDH2 mutations are identified in approximately half of cases (9); negativity for CKAE1/AE3 ruled out epithelial component; and a proliferation index by Ki-67 close to 1%, along with absence of necrosis and atypical mitotic figures, defined the benign nature of the lesion (1). The integration of these morphological and immunohistochemical findings, together with clinical and imaging correlation, allowed a solid definitive diagnosis. Additionally, the imaging features of the present case, namely relative preservation of the mandibular condyle and predominance of erosive involvement of the glenoid roof, are consistent with synovial chondromatosis and discordant with the typical findings of synovial chondrosarcoma, which tends to be centered on the condyle and to spare the glenoid fossa roof (10).

Two anatomical findings of the case deserve particular attention. The first is bicompartamental involvement, supradiscal and infradiscal, which constitutes an atypical presentation of TMJ SC, where isolated involvement of the upper compartment is usual (3, 4). The presence of loose bodies in both spaces suggests a long-standing disease and requires consideration of a possible previous or concomitant alteration of the integrity of the articular disc (3). The second, and probably most relevant from the surgical standpoint, is the dehiscence of the glenoid fossa roof toward the middle cranial fossa, identified intraoperatively. Although erosion and thinning of the glenoid roof have been described in advanced forms of TMJ SC (4, 7), frank dehiscence with potential intracranial communication is an infrequent finding with critical clinical implications (7, 8): it conditions the extent of dissection, requires extreme caution in the medial recess, and should prompt, in future patients with imaging suspicion of glenoid fossa roof erosion, the request of thin coronal CT

sections with multiplanar reconstruction as the imaging modality of choice for the assessment of the bony component (7). Magnetic resonance imaging, although it does not allow direct evaluation of cortical bone, is complementary by providing information about the articular disc, the synovium, and the potential extension to extra-articular soft tissues. This finding also justifies, in this scenario, the choice of an open preauricular approach over arthroscopic alternatives, since the latter would have significantly limited the assessment of the glenoid roof and the intraoperative modulation of surgical conduct (4).

The systematic indication of synovectomy associated with the excision of loose bodies has traditionally been recommended with the aim of reducing the risk of recurrence (4, 6). However, this principle is based on the metaplastic activity of the synovium, which in Milgram stage III is already inactive (5). In the present case, the decision not to complete formal synovectomy was based on the dehiscence of the glenoid roof and on the need to avoid potential dural injury. This conduct is consistent with the surgical principle of modulating the aggressiveness of resection according to local anatomical conditions and the histopathological stage of the disease (4, 6). The persistence of residual loose bodies in the medial recess documented in the postoperative CT was not associated, during the eight-month follow-up, with clinical manifestations, signs of articular progression or indications of reactivation, supporting the relevance of the conduct adopted in this specific scenario.

It is appropriate, however, to underline the limitations of this work. The eight-month follow-up, although it allows documentation of satisfactory symptomatic and functional resolution in the early postoperative period, is insufficient to draw definitive conclusions about the risk of long-term recurrence, given that synovial chondromatosis can recur even years after surgery (3, 4).

Although preoperative measurement of maximum mouth opening was available (30 mm), allowing documentation of objective functional improvement after surgery, measurements of lateralities and protrusion were not recorded preoperatively, which partially limits the comparative global functional evaluation. Additionally, preoperative magnetic resonance imaging was not available, a limitation inherent to the healthcare setting in which the patient was treated. Moreover, the preoperative CT study did not include thin coronal sections centered on the glenoid roof, an imaging technique that would probably have allowed anticipation of the bony thinning identified during surgery. Likewise, no new tomographic study was performed at eight months; postoperative imaging control of the present report was limited to the immediate postoperative study, with new controls scheduled for subsequent follow-ups. The patient continues clinical and radiological follow-up every six months, in agreement with current literature recommendations given the rarely reported risk of local recurrence and malignant transformation in chronic lesions (1, 4).

CONCLUSIONS

Synovial chondromatosis of the temporomandibular joint is an uncommon entity whose diagnosis requires a high index of clinical suspicion and, fundamentally, histopathological confirmation complemented by immunohistochemical study, especially when factors such as atypical age of presentation or mild cytological atypia are present, in which case the differential diagnosis of synovial chondrosarcoma must be rigorously ruled out.

The present case illustrates an unusual presentation of the disease, with bicompartamental involvement and dehiscence of the glenoid fossa roof toward the middle cranial fossa in a seventh-decade patient, in which local anatomical conditions required modulation of surgical management. Subtotal excision of loose bodies,

without formal synovectomy, allowed in this specific scenario satisfactory symptomatic and functional resolution in the early postoperative period, without clinical or radiological signs of reactivation at eight months of follow-up.

These results should be interpreted with caution and do not allow generalizing the indication of a subtotal approach to other cases, given the short duration of follow-up. Prolonged clinical and radiological follow-up is required to confirm the stability of residual bodies and to rule out long-term recurrence, as well as series with larger numbers of cases and extended follow-up to specify the role of synovectomy and the criteria for subtotal excision in advanced TMJ synovial chondromatosis, particularly when complex anatomical findings such as glenoid roof dehiscence are present.

DECLARATIONS

ETHICAL CONSIDERATIONS: This case report was conducted in accordance with the principles of the Declaration of Helsinki of the World Medical Association. Written informed consent was obtained from the patient for the publication of this case report, its associated clinical data and images. All identifiable data have been anonymized.

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