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# Innovative Surgical Management of the Synovial Chondromatosis of Temporo-Mandibular Joints: Highly Conservative Surgical Technique

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**Abstract:** Synovial chondromatosis (SC) is an uncommon disease characterized by a benign nodular cartilaginous proliferation arising from the joint synovium, bursae, or tendon sheaths. Although the temporomandibular joint is rarely affected by neoplastic lesions, SC is the most common neoplastic lesion of this joint. The treatment of this disease consists in the extraoral surgery with a wide removal of the lesion; in this study, the authors described a more conservative intraoral surgical approach. Patient with SC of temporomandibular joint typically refer a limitation in the mouth opening, together with a persistent not physiological mandibular protrusion and an appearance of a neof ormation located at the right preauricular region: the authors reported 1 scholar patient. After biopsy of the neof ormation, confirming the synovial chondromatosis, the patient underwent thus to the surgical excision of the tumor, via authors' conservative transoral approach, to facilitate the enucleation of the neof ormation. The mass fully involved the pterygo-maxillary fossa with involvement of the parotid lodge and of the right TMJ: this multifocal extension suggested for a trans-oral surgical procedure, in the light of the suspicion of a possible malignant nature of the neoplasm. Our intraoral conservative approach to surgery is aimed to reduce the presence of unaesthetic scars in preauricular and facial regions, with surgical results undoubtedly comparable to the traditional surgical techniques much more aggressive. Our technique could

be a valid, alternative, and safe approach to treat this rare and complex kind of oncological disease.

**Key Words:** Conservative surgery, oncological surgery, synovial chondromatosis, temporomandibular joint neoplasm

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Synovial chondromatosis (SC) is an uncommon disease who was described for the first time in the 16th century by Ambroise Paré.<sup>1</sup> The World Health Organization<sup>2</sup> defines the SC as a benign nodular cartilaginous proliferation arising from the joint synovium, bursae, or tendon sheaths.

The pathogenesis of SC is characterized by the multiple cartilaginous nodules, or bodies, formed in the synovial membrane. These formations may be either covered (attached) or uncovered (detached) by the synovial lining cells and fibrous tissues.<sup>2</sup> Three phases of the SC have been described<sup>3</sup>: active intrasynovial disease with nodules but no calcifications/intrarticular bodies; synovitis with osteochondral nodules in the synovial membrane and loose bodies within the joint; and multiple loose bodies remain but synovitis is quiescent. The malignant transformation of primary SC to synovial chondrosarcoma is reported to be a rare event with an estimated incidence of 1% to 5%.<sup>4–6</sup>

Synovial chondromatosis usually manifests between the third and the fifth decades of life,<sup>7</sup> with men being affected 2 or 4 times more frequently than women.<sup>8</sup> The diagnosis is made by computerized tomography (CT) and magnetic resonance imaging (MRI).<sup>9</sup> The correlation between histological and radiological diagnosis is of significant importance to correctly diagnose the benign or the malignant entity of the lesion.

Clinical symptoms typically include pain, swelling, and restricted joint movement.<sup>10</sup>

The knee is the most commonly affected joint (70% of the patients)<sup>5</sup> followed by the hip (20% of the patients), the shoulder, the elbow, the ankle, and the wrist (less than 10% of the patients).<sup>11</sup> The symptoms are often deceitful at disease onset and are gradually progressive, although rare spontaneous regression has been reported.<sup>12</sup> Treatment of SC usually consists of surgical removal of the intrarticular bodies with or without synovectomy, but local recurrence is not uncommon, occurring in up to 3% to 23% of the patients.<sup>5,13</sup>

Although the temporomandibular joint (TMJ) represents one of those sites less affected by tumors or tumor-like lesions, SC is the most common neoplastic lesion of this joint.<sup>14</sup> Temporomandibular joint SC patients are divided into primary SC, without identifiable etiological factors, and secondary SC with a known etiology.<sup>15</sup> Secondary patients are often thought to be related to previous trauma, repetitive microtrauma, or degenerative arthritis. Primary

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patients cannot be associated with any supposed etiological factors and are considered to be more aggressive.<sup>15</sup>

According to a systemic search in the National Library of Medicine's PubMed Database, 155 patients were described in 103 publications;<sup>16</sup> women are affected more than men, with an average age of 46-year old.<sup>16</sup>

Most of the studies described open surgery interventions;<sup>17</sup> synovectomy was the most frequent procedure associated with loose body removal from the joint space, often accompanied by diskectomy and, less frequently, by condilectomy.<sup>15</sup>

In the present patient, we opted for an intraoral surgical approach instead of the classic transcuteaneous approach since this technique can reduce the morbidity of the surgical site, as well as can avoid potential aesthetic damages, without reducing the effectiveness of the surgery.

## METHODS

### Maxillofacial Units

The reported surgical technique has been performed in close collaboration between the Unit of Maxillofacial Surgery of the Calabro dental Clinic (Crotona, Italy), the Biomedical Section of the Tecnologica Research Institute (Crotona, Italy), the Advanced Diagnostic Labs of the Marelli Hospital (Crotona, Italy), and the National Tumor Institute "Fondazione G. Pascale" (Naples, Italy).

Ethical approval was given by the ethical committee of National Tumor Institute "Fondazione G. Pascale" (Naples, Italy).

A written informed consent was obtained from the patient by using a specific form. The study followed the "ethical principles for medical research involving human subjects" of the last modification of Helsinki Declaration. The study was conducted in accordance with Italian laws and regulations.

### Patient Chosen to Be Reported

We used this technique on the rare patients of SC of the TMJ; we found suitable for scientific purpose one of them. A 62-year-old woman referred a limitation in the mouth opening and of the mandibular lateral movements, together with a persistent not physiological mandibular protrusion: this condition was constantly increased from about 5 months; furthermore, in the last month, the patient reported the appearance of a neoformation located at the right preauricular region associated with a swelling of tense-elastic consistency, and painful symptoms at palpation.

Physical examination revealed the inability to open the mouth, with a maximum interincisive distance of 20mm, and the severe limitation of any lateral movement. Intraoral palpation revealed a right parotid neoformation of approximately 50mm; the right parotid region appeared covered with unaltered skin, and the lump was not displaceable at palpation.

Computerized tomography of the neck and the chest and MRI of the maxillofacial bones were performed revealing an expansive lesion of 46 × 54 mm size, with intralésion septa and a liquid content associated with diffuse calcifications (Figs. 1 and 2). The side wall of the maxillary right sinus appeared eroded in a large area in contact with the lesion. Furthermore, lumbo-sacral osteoarthritis events were present. It was therefore decided to carry out a biopsy of the neoformation which resulted composed by hyperplastic cartilaginous tissue with marked cellular *atipia*.

## RESULTS

### The Surgery

The patient underwent then the surgical excision of the mass via a conservative and innovative trans-oral approach, to facilitate the



**FIGURE 1.** Coronal plane. (A) Computerized tomography examination. Preoperative image of a neoformation developed in the right pterygo-maxillary fossa. (B) NMR examination with contrast medium. Preoperative image of a neoformation developed in the right pterygo-maxillary fossa; the lesion penetrates also in the parotid loggia.

enucleation of the numerous components of the neoformation (Fig. 3).

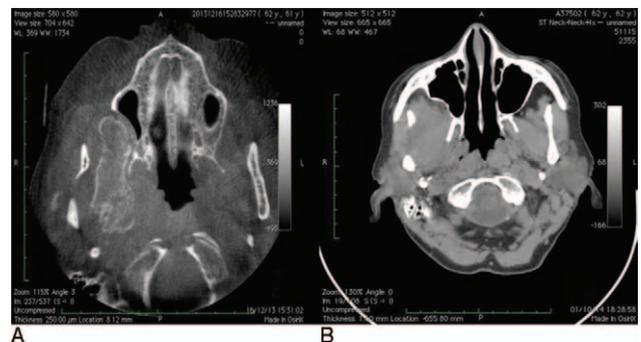
### The Histology

Finally, the histological examination revealed that the lesion was compatible with a SC (Fig. 4); the biopsy specimen resulted negative for biomarkers of soft tissue malignancies, such as S100, Actin 1A4, and CD34.<sup>18-20</sup>

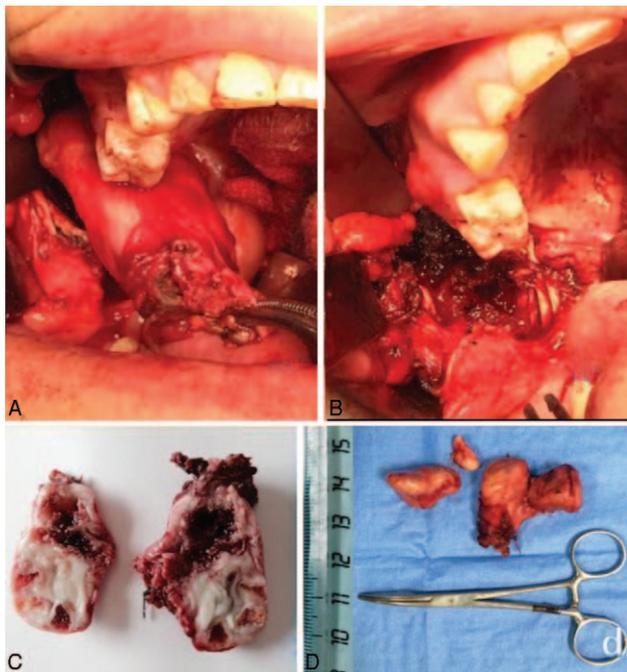
The mass involved the pterygo-maxillary fossa with involvement of the parotid lodge and of the right TMJ; this multifocal extension suggested for a trans-oral surgical procedure, in the light of the suspicion of a possible malignant nature of the neoplasm. By means of CT and MRI images it was possible to identify a clear cleavage plane between the mass and the surrounding tissues: due to this approach, we were able to detach the neoformation from the lateral wall of the maxillary sinus and single it out from the parotid lodge. Sigmoid incisures allowed separating the neoplasm from the TMJ capsule.

## DISCUSSION

Synovial chondromatosis is a rare, benign process that typically affects large joints in young adults. Although it commonly involves the knee,<sup>5</sup> it may occur in the TMJ. The disease is characterized by the development of cartilaginous nodules within the synovial space from the synovial connective tissue matrix;<sup>21</sup> the nodules subsequently degraded, detached, and become fluctuating intrarticular bodies tending to calcify in the joint space. The involved joint space



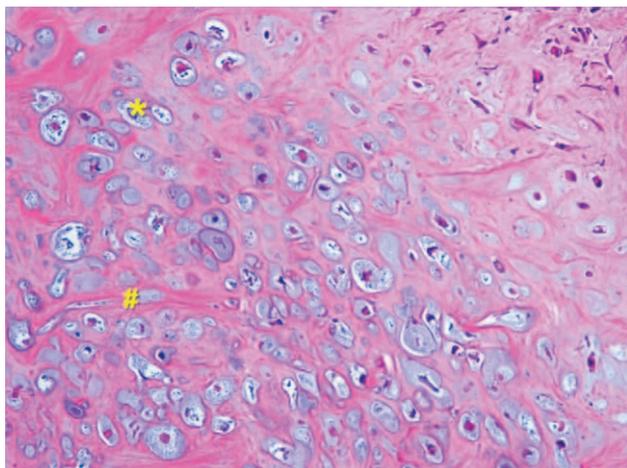
**FIGURE 2.** Axial plane. (A) CT examination. Preoperative image of a neoformation developed in the right pterygo-maxillary fossa. (B) CT examination. Postoperative image: no sign of lesion is clinically evident. CT, computerized tomography.



**FIGURE 3.** (A, B) The composite figure shows the intraoperative phases with the enucleation of the lesion by using the intraoral approach. (C, D) The intraoperative pieces have been processed and analyzed by the histopathology section.

may also become widened with possible articular erosions, eventually leading to secondary osteoarthritis.

Synovial chondromatosis of other joints has been reported to occur twice as often in males as in females, with a mean age of onset in the fifth decade.<sup>22</sup> In the patient of TMJ, it occurs more often in females by a ratio of 4:1, and is usually located on the right side (right to left ratio of 4:1).<sup>23</sup>



**FIGURE 4.** Synovial chondromatosis of temporomandibular joint is a rare form characterized by the presence of nodules of metaplastic cartilage in the synovium of this joint. The presenting symptoms include pain and limitation of the joint movement. The cartilage nodules may be embedded within the synovium covered by a layer of flattened synovial cells or lie freely within the joint cavity. The lesion shows clusters of chondrocytes (°) separated by solid chondroid matrix (#). The chondrocytes frequently show moderate-to-severe atypia. Taken out of context, the histologic features may be mistaken for Grade 1 chondrosarcoma.

In 1933, Axhausen<sup>24</sup> described the first patient of SC affecting the TMJ. The etiology remains unknown, but generally a history of trauma and recent inflammations is often found in patients.<sup>24</sup>

Histologically, SC may be divided into 3 stages of development: metaplasia found in the synovial membrane without the presence of detached particles, metaplasia found in the synovial membrane with the presence of detached particles, and presence of detached particles, which may vary in size from less than 1 mm to greater than 10 mm.<sup>25</sup>

In the patient of larger joints, the classical triad of signs and symptoms is represented by restricted joint range of motion (65% of the patients), pain (57% of the patients), swelling (46.5% of the patients),<sup>16</sup> which are also the main signs and symptoms identified in patients of SC of the TMJ.<sup>26</sup> These features are often nonspecific, overlapping those of other TMJ diseases.<sup>27</sup> In case of absence of physical signs, the development of nonspecific pain and headaches can lead to a delay in diagnosis or to a misdiagnosis of other more common causes of headaches.<sup>28</sup> Data on the duration of the symptoms before diagnosis and intervention show that in the 80% of the patients reported in the literature, symptoms lasted for more than 2 years.<sup>16</sup>

Advanced imaging techniques, such as MRI and CT, are now being used with great success to evaluate TMJ disorders,<sup>27</sup> helping to depict joint changes and the presence of loose bodies, suggesting the diagnosis of SC.<sup>29</sup> Progresses in CT and MRI have improved the ability to delineate TMJ disease markedly, particularly with the use of sagittal and coronal section imaging. Magnetic resonance imaging may be useful for depicting the nodules in the early stages of formation, before ossification,<sup>24</sup> and for planning surgery in the early stages. Noyek et al<sup>22</sup> pointed out the radiologic features of SC in the TMJ: widening of the joint space, limitation of motion, irregularity of the joint surface, presence of calcified loose bodies (cartilage), and sclerosis or hyperostosis (overgrowth) of the glenoid fossa and mandibular condyle. These radiologic features, however, are also commonly seen in osteoarthritis involving the TMJ, except for the presence of calcified loose bodies.<sup>30</sup> For this reason, differential diagnosis is also very important. Intra-articular temporomandibular pain most commonly is due to degenerative osteoarthritis, a typical progressive disease, osteosarcoma and chondrosarcoma, the most frequent, which are malignant diseases that can arise within the TMJ.<sup>31</sup> Other causes of loose bodies within a joint include osteochondritis dissecans, the most common one,<sup>32</sup> intracapsular fracture, avascular necrosis, tuberculous or pyogenic arthritis, rheumatoid arthritis, and neutrophilic arthritis.<sup>32,33</sup> The surgery has always been recommended as therapy of choice,<sup>3,28</sup> but some authors advocate less invasive techniques, such as arthroscopy and 2-needle arthrocentesis, to remove the loose bodies from the joint space.<sup>31</sup> Many authors suggested that CT, MRI imaging, and arthroscopy can facilitate the clinical diagnosis of synovial chondromatosis.<sup>25,34,35</sup> Nevertheless, a definitive diagnosis can be made only by histological examination.<sup>27</sup> In the reported case, the immunohistochemical analysis was negative for the following markers of malignancy: CD34, S100, and Actin 1A4;<sup>18–20</sup> this laboratory analysis was leading to confirm the suspect of the diagnosis of chondromatosis, erasing in this way the initial doubts derived from the analysis of the radiological findings.

According to the latest suggestions, TMJ arthroscopy has no advantages over arthrocentesis in terms of efficacy, and none over open surgery in terms of postsurgical course.<sup>36,37</sup> In many patients, additional procedures, such as total synovectomy, diskectomy and condylectomy, were performed for loose bodies' removal alone; the choice of the surgical procedure should be based on the stage of the disease.<sup>25,28</sup> In patients of an advanced stage of the disease, characterized by degeneration and calcification of the loose bodies and by a nonproliferative inactive synovial membrane, aggressive

surgery with synovectomy might not be necessary.<sup>16</sup> Since literature data do not provide useful information on the correlation between the stage of the disease and the type of surgical intervention, the experience of the surgeon is the main determinant;<sup>16</sup> moreover, the intraoral condition should be carefully evaluated so to avoid intraoral infections or diseases.<sup>38</sup>

Once the loose bodies have been removed, the recurrence rate of SC appears to be very low; only 1 patient has been documented in the literature.<sup>39</sup> This may suggest a spontaneous inactivation of the metaplastic process in most patients and, at the same time, that surgery can be considered a radical and definitive treatment.<sup>40,41</sup>

In conclusion, a correct clinical and instrumental approach, together with a multidisciplinary evaluation (pathological, oncological, and surgical), allowed performing a radical and clean surgical procedure. The marked cellular atypia emerged from the first biopsy, closely linked to hypothetical doubts of pulmonary metastases, has been clearly investigated by means of the reported postsurgery histological examinations. Three months after the surgery, lung and maxillofacial TC control showed a reduction of the lesion and the absence of residual local disease.

## Take-Home Message

Due to our intraoral approach, the patient will avoid unaesthetic scars<sup>42</sup> in the preauricular and facial regions without otherwise reducing the effectiveness of surgery; we can obtain that the patient regains a correct mouth opening with relative mandibular protrusion and lateral movements, close to normality. Our technique could represent a safe alternative approach<sup>43</sup> aimed to treat this rare oncological pathology.

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