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A rare case of chondroblastoma of the temporomandibular joint: A case report

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ABSTRACT

Chondroblastoma is a rare and benign bone tumor that can occur in the craniofacial region. It may be difficult to diagnose using imaging alone; histological analyses and even immunohistochemical analyses are often required. This article reports on a rare case of chondroblastoma of the temporomandibular joint. Its diagnosis was hindered by its similarity to other tumors in both imaging exams and histological analyses. It was definitively treated through the total excision of the tumor.

1. Introduction

Chondroblastoma was first established in 1931 by Codman, who reported nine cases of a growth that he referred to as "a giant cell chondromatous tumor of the epiphysis" [1]. In 1942, Jaffe and Lichtenstein distinguished chondroblastoma from giant cell tumors and created the term "benign chondroblastoma." They hypothesized that this tumor may originate from remains of the fetal chondroid skeleton [2]. It is a benign cartilaginous neoplasm that represents approximately 1% of all primary tumors affecting the epiphyses of long bones, particularly in the proximal tibia or humerus, as well as in the distal femur. It rarely forms in the region involving the mandibular condyle [3–6]. The rarity of this tumor increases exponentially when it is a case of chondroblastoma that is extraosseous and in the region involving the temporomandibular joint (TMJ) [7]. The following report describes one of the few cases of extraosseous chondroblastoma on the TMJ. It was successfully treated through total excision of the tumor.

1.1. Case report

A 46-year-old patient reported that 3 years before the discovery of chondroblastoma, he started pain in the left temporomandibular region. As a previous medical history it presented only discoid lupus but already in follow-up with dermatologist, without any history of trauma or surgery or some internal disorder in temporomandibular joint (TMJ). As pre-auricular pains increased in intensity as the days went by, he sought medical treatment. The initial medical management was by means of analgesics for a period of 10 days. The pain subsided and the patient followed without further care.

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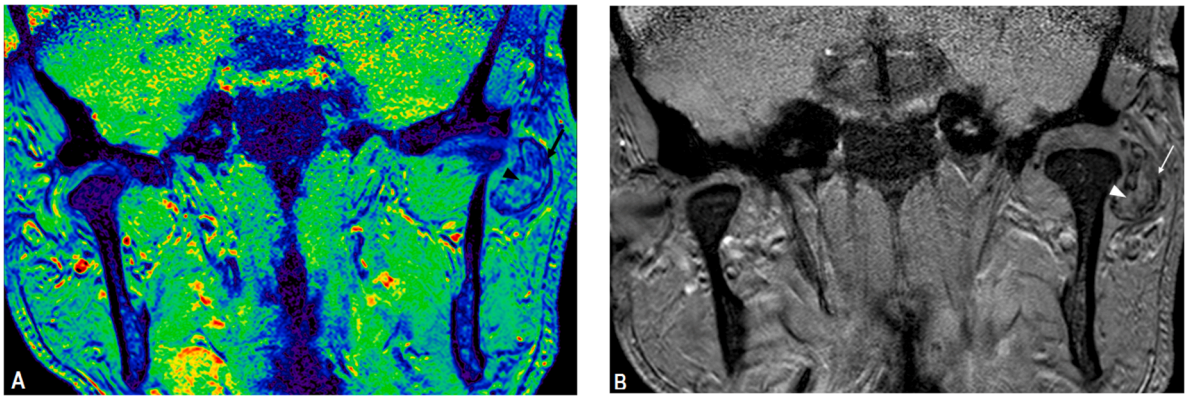

Fig. 1. A and 1B - Magnetic resonance imaging of the left TMJ in a T1-weighted coronal view, showing an oval-shaped lesion, surrounded by a low-signal halo (arrow) and presenting a heterogeneous component compatible with high-signal calcifications (arrowhead). (A) Post-processed MRI by dynamic color enhancement.

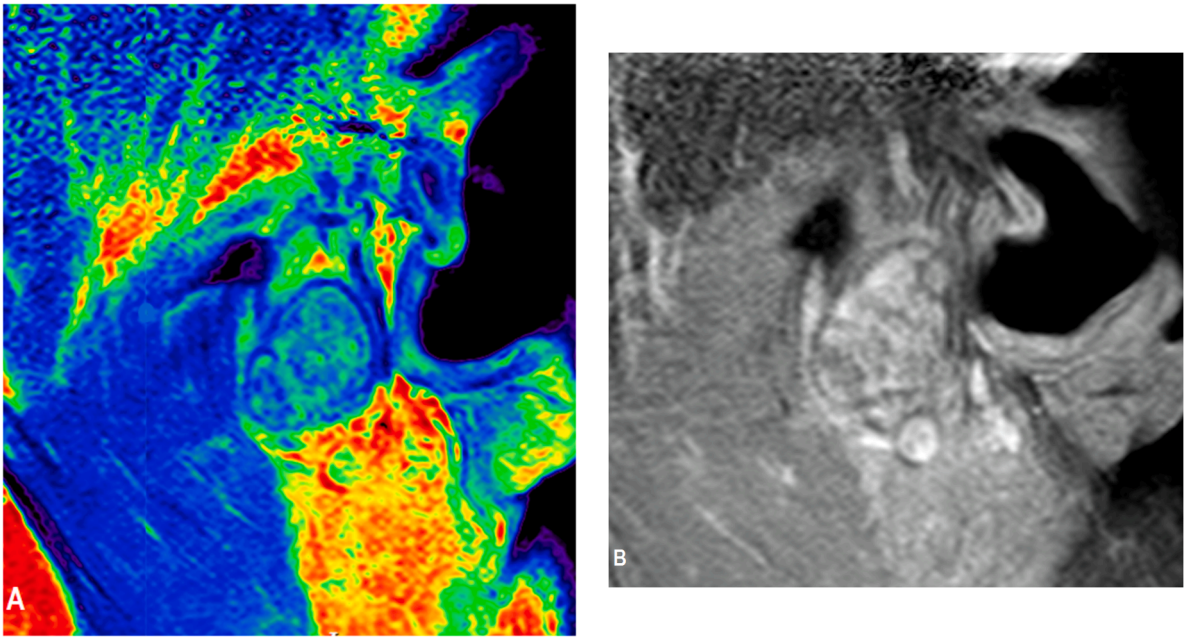

Fig. 2. A and B – Magnetic resonance imaging of the left TMJ in a T1-weighted sagittal view, showing an oval-shaped lesion, surrounded by a low-signal halo and presenting a heterogeneous component compatible with high-signal calcifications. (A) Post-processed MRI by dynamic color enhancement.

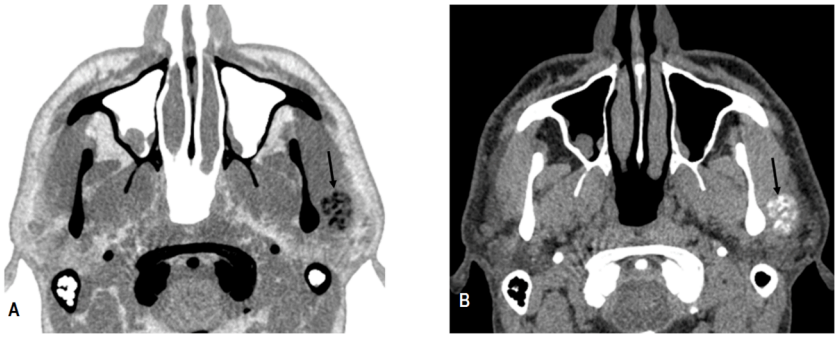

Fig. 3. (A/B) Computed tomography in axial view. The lesion seemed internally mixed lateral to the left condylar process and exhibited a discretely hypodense halo (arrow).

Two years later, the patient returned with pain that had spread to the left auricle and which was associated with increased volume at the site. He sought care from an otolaryngologist, who ordered an MRI and CT of the region. The MRI revealed prominent bicompartmental joint effusion with thickening and diffuse synovial impregnation by contrast both more prominent to the left side, associated with a clearly defined lesion. The lesion was oval-shaped, surrounded by a hypointense halo sign. The inner portion exhibited a heterogenicity compatible with calcifications with a high signal in sequences sensitive to liquids. The dimensions were $2.1 \times 1.0 \times 1.7$ cm (Fig. 1a, b, 2a, and 2b). In the CT, the lesion seemed internally mixed lateral to the left condylar process and

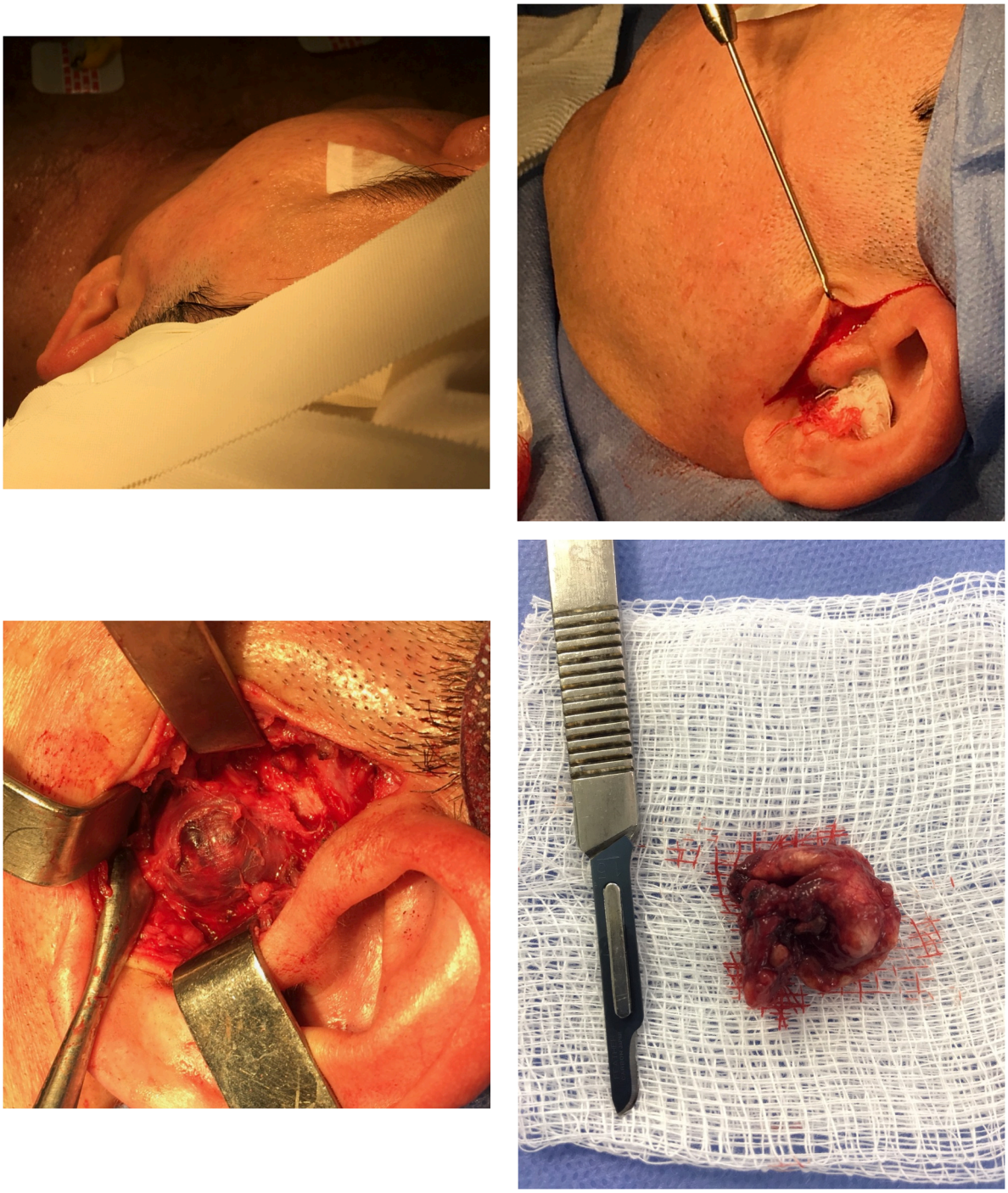

Fig. 4. a – Top-inferior view showing the lateral bulging of the fluid by the lesion, b – demarcation of pre-auricular access for tumor removal, c – Chondroblastoma in dissection. In this image we can also perceive the fossa articular superiormente lesion, Figura 4d – lesion removed. Note the large dimension of chondroblastoma.

exhibited a discretely hypodense halo (Fig. 3).

The patient was then sent to an oral and maxillofacial surgeon and underwent surgery with general anesthesia on September 29, 2017, to completely excise the lesion (Fig. 4a, b, 4c and 4d). The tumor was excised using the preauricular approach due to its dimensions and was then sent for histopathological analysis. The histological analysis revealed areas with chondral calcification, as well as areas that were cartilaginous in nature mixed with synovial tissue with polygonal stromal cells and multinucleated giant cells (Fig. 5). The clinical and histological findings were insufficient to establish a diagnosis, since they did not allow for the exclusion of other, histologically similar tumors such as synovial chondromatosis or pigmented villonodular synovitis with cartilaginous metaplasia. For this reason, an immunohistochemical analysis was performed. It revealed fragments of hypercellular neoplasm with multinucleated giant cells (Fig. 6a) and fine linear calcifications surrounding the stromal cells in a "chicken wire" or lace-like pattern. These fragments were associated with the formation of immature chondroid material (Fig. 6b). An immunohistochemical stain for S-100 protein (Fig. 6c) was positive in neoplastic cells inside and outside the cartilaginous matrix, showing its chondroid differentiation, making the diagnosis compatible with chondroblastoma.

The patient is currently receiving outpatient follow-up care from physicians specializing in oral and maxillofacial surgery, dermatology, and rheumatology. Up to now, he has been following up on 18 months and He has had no functional complaints regarding his TMJ, and there have been no radiographic findings suggestive of chondroblastoma recurrence.

2. Discussion

Since they were was first classified as a bone and joint tumor and subsequently defined as a benign tumor by Jaffe and Lichtenstein in 1942 [2,8], chondroblastomas have represented 1% of all bone tumors [9]. Chondroblastomas most commonly occur in the bones of the limbs, such as the distal femur and the proximal tibia or humerus; there have been few reports of chondroblastoma of the mandibular condyle, since this tumor is rarely found in the craniofacial region [3–6,10]. In the literature, this tumor in the craniomaxillofacial region is found much more frequently in the temporal bone. To date, only 13 cases of chondroblastoma in TMJ have been reported in english literature [11–23,25].

Pain is the most important symptom in chondroblastoma of the limbs [5]. Tumors that occur in the TMJ may exhibit symptoms similar to those associated with other temporomandibular disorders. These symptoms include progressive functional restrictions, focal pain, noise in the jaw when biting, and limited range of motion when opening or closing the mouth [11,15,22]. In this case, the patient reported local pain, limited jaw function, and increased volume of the site.

Imaging-based criteria for diagnosing chondroblastoma of the TMJ have not been standardized or clearly defined, likely because of the rarity of this type of tumor. Radiographically, the tumor has been represented by resorptive defects or by condylar enlargements

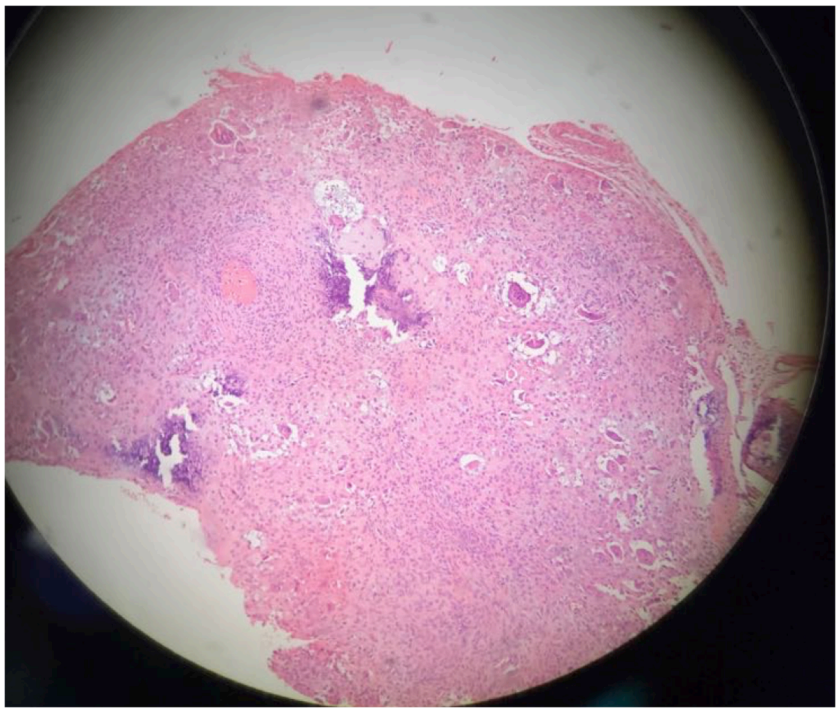

Fig. 5. Histopathological images of the mass (H&E stain; original magnification The histological analysis revealed areas with chondral calcification, as well as areas that were cartilaginous in nature mixed with synovial tissue with polygonal stromal cells and multinucleated giant cells besides old hemorrhage, and calcification.

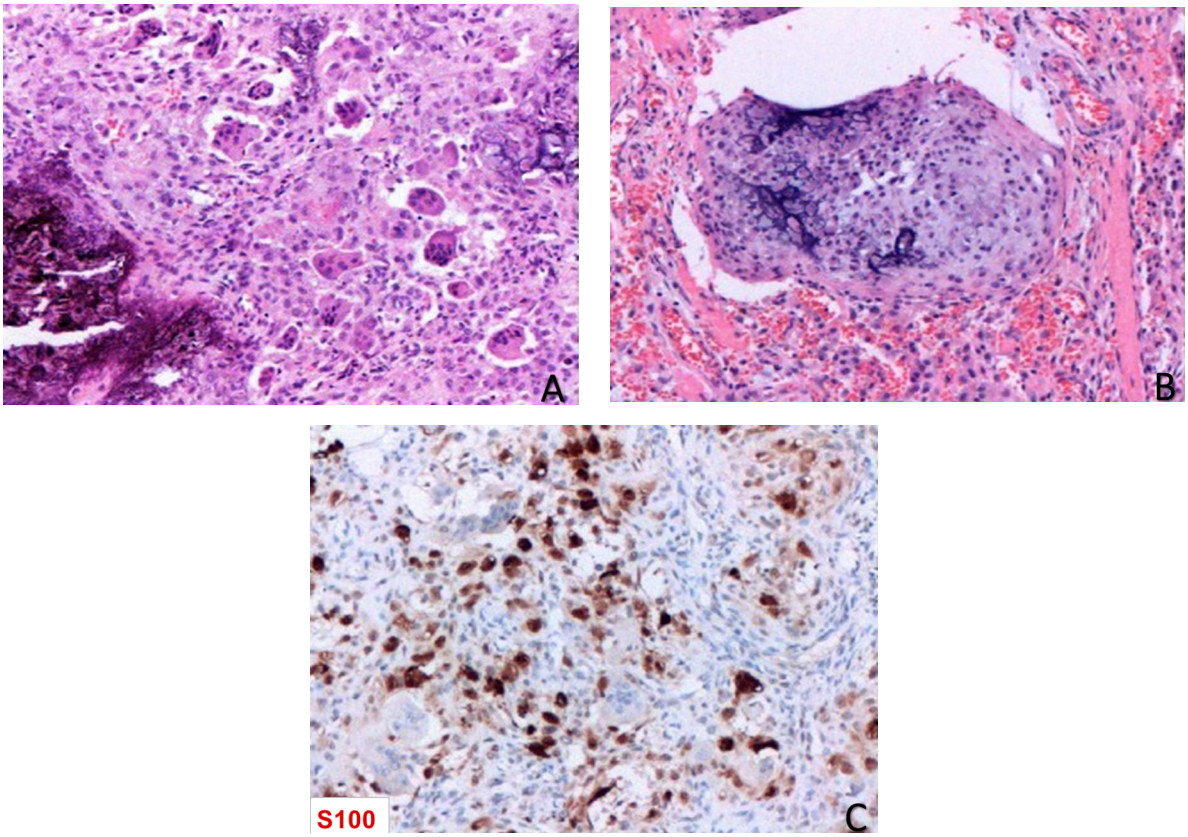

Fig. 6. a- Immunohistochemical analysis revealed fragments of hypercellular neoplasm with multinucleated giant cells. b - fine linear calcifications surrounding the stromal cells in a "chicken wire" or lace-like pattern. These fragments were associated with the formation of immature chondroid material. c - An immunohistochemical stain for S-100 protein.

associated with thinning of the cortex [10]. Bui et al. [23] used CT and MRI and found images compatible with mass expansion of the condyle to the branch, lithic irregularities, substantial radiolucency with a thin sclerotic margin, condylar enlargement, and destruction of the squamous portion of the temporal bone, the glenoid fossa and the zygomatic arch. The MRI revealed joint effusion with signs of synovitis on the left side associated with a clearly defined lesion. The lesion was oval-shaped, surrounded by a hypointense halo sign. The inner portion exhibited a heterogenicity compatible with calcifications with a high signal in sequences sensitive to liquids. In the CT, the lesion seemed internally mixed lateral to the left condylar process and exhibited a discretely hypodense halo. However, despite the large quantity of information that can be obtained from imaging exams, the precise differential diagnosis of this tumor relative to other temporomandibular conditions (such as chondrosarcoma, osteosarcoma, malignant fibrous histiocytoma, chondroma, giant-cell tumor of bone, aneurysmal bone cyst, and eosinophilic granuloma) is difficult based on clinical and imaging exams alone; histopathological analyses are also required [7,10,17,23].

In this case, the histological findings included chondroid areas exhibiting a relatively acellular amorphous matrix occasionally containing round and oval-shaped chondrocyte cells inside empty spaces, as well as a dense concentration of small polygonal cells or round chondroblasts within chondroid foci. Few cells in mitosis were observed. Multiloculated giant cells have also been seen in some areas of cells but are not as numerous as in giant cell tumors with no calcifications [7,10]. However, despite the similarity of these histological findings to those in the literature [7,10], they were insufficient to make the diagnosis. They did not allow for the exclusion of other tumors such as synovial chondromatosis or pigmented villonodular synovitis with cartilaginous metaplasia.

The presence of the S-100 protein in tumor cells differentiates chondroblastoma from other pathological processes with histological similarities, since the immunoreactivity of this protein has been shown to be associated with the formation of chondroid tissue, and chondroblastomas exhibit strong immunoreactivity to S-100 [7,10,23,24]. Thus, after the immunohistochemical analysis, a diagnosis compatible with chondroblastoma was possible.

Though benign, chondroblastoma can cause osteolysis because they exhibit aggressive focal characteristics, including bone invasion. Questions remain regarding the use of surgical treatment for this type of tumor. Bui et al. [23] considered thirteen cases in which the treatment reported included curettage, resection, or excision of the tumor. Of these cases, there was recurrence in only one, whose treatment was initially done by curettage. Recurrence occurred 3 years after treatment, but it was successfully treated by the repetition of the same procedure and was followed for another three and a half years, without recurrence of the lesion. In two of the reports they considered, the treatment was not documented. In none of the cases treated with resection did the tumor return. The

follow-up periods for the patients considered ranged from 12 months to 13 years [25].

This type of tumor can be treated with curettage, simple excision (considering the type of lesion and surrounding anatomical structures), total excision, or surgery combined with radiation; however, most studies suggest total resection as the most effective method for reducing the possibility of recurrence.

The professionals responsible for these cases should therefore carefully consider what constitutes a successful treatment, as well as the benefits of complete tumor excision, which depend on the degree of invasion of the surrounding anatomical structures. In this case, in which the tumor had not invaded the bone structures, it was treated using block excision. The patient experienced no postoperative infections, reported no major complaints, and has continued outpatient follow-up care with physicians specializing in oral and maxillofacial surgery, dermatology, and rheumatology. There have been no reports of functional problems in the TMJ, nor any imaging findings suggestive of tumor recurrence.

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