Chondrosarcoma of the Temporomandibular Joint: A Case Report in a Child

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This article reports a rare case of a temporomandibular joint (TMJ) chondrosarcoma in a child. Chondrosarcoma is a malignant cartilaginous neoplasm that resembles synovial chondromatosis. In the head and neck region, chondrosarcoma is uncommon, corresponding to 6.4% to 12% of all reported cases. The majority of patients with chondrosarcoma are in the third to fourth decades of life. A Pubmed search showed that 20 TMJ chondrosarcoma cases had been reported up to 2008. The present case was of an 11-year-old girl referred to an Oral Disease Center and presenting with a preauricular swelling on the right side and normal ENT evaluation. The patient was healthy. Discrete pain and mild limitation of mouth opening were observed. A panoramic radiograph as well as computed tomography (CT), ultrasound, and magnetic resonance imaging (MRI) revealed an osteolytic lesion in the right TMJ. The skull base and adjacent spaces were preserved but adjacent anatomic structures were displaced. After an incisional biopsy, the patient underwent high condylectomy. Microscopic findings showed a tumor exhibiting cartilaginous tissue proliferation with celullar pleomorphism, nuclear hyperchromasia, and mixoid changes in the matrix. The immunohistochemical analysis of the expression of Ki-67 and Cyclin B1 proteins (cellular proliferation markers) revealed a very low proliferative cell index. The 3.5 years of clinical and imaging follow-up have shown no evidence of recurrence or metastasis, but signs of myofascial disorders could be observed. It is concluded that cartilaginous lesions in the jaws must be regarded with suspicion, since benign and malignant lesions may show similar clinical features. This case emphasized the importance of interdisciplinary approaches to minimize the possibility of misdiagnosis. J OROFAC PAIN 2009;23:275–281

Key words: chondrosarcoma, malignant neoplasm, synovial, temporomandibular joint

Chondrosarcoma is a malignant cartilaginous neoplasm arising from normal chondroid tissues or from embryogenic cartilaginous rest.[1] The most common sites are the pelvis, ribs, femur, and humerus.[2,3] Based on microscopic criteria, the neoplasm can be divided into two major types: conventional chondrosarcoma and chondrosarcoma variants.[4] Conventional chondrosarcoma can be graded as I (low), II (intermediate), and III (high). Grading is subjective and based on relative degrees of cellularity, atypia, and pleomorphism. The variant type is further divided into four subtypes: clear cell, myxoid, dedifferentiated, and mesenchymal.[2,4]

Table 1 The 20 TMJ Chondrosarcoma Cases Published from 1960 to 2008*

Reference no. and author	Sex	Age (y)	Symptoms duration (mo)	Chief complaint†	Imaging features	Surgical treatment	Irradiation treatment	Recurrence follow-up (mo)
3. Sesenna et al (1997)	F	60	12	Swelling	Mass from condyle to infratemporal fossa with calcification	Yes	No	60
8. Mostafapour and Futran (2000)	F	31	96	Swelling	Mass extending into the ipsilateral pterygoid space	Yes	Yes	Lost contact
8. Mostafapour and Futran (2000)	F	52	18	Swelling	Mass with mineral density on the temporal bone	Yes	No	12 recurrence
9. Merrill et al (1997)	F	50	36	Mouth opening and mastication difficulty	Circumscribed radiolucent-radiopaque mass surrounding condyle	Yes	No	18 no recurrence
10. Gingrass (1954)	F	46	12	Swelling and pain	Slight articular space widening, subcortical sclerosis	Yes	No	Not reported
11. Lanier and Wilkinson (1971)	F	48	24	Swelling and pain	Condyle resorption	Yes	No	Few months
12. Richter et al (1974)	M	75	10	Swelling and pain	Articular space widening	Yes	No	12
13. Tullio and D'Errico (1974)	F	17	8	Swelling	Condyle resorption	Yes	No	Not reported
14. Nortjé et al (1976)	M	40	6	Dull pain and swelling	Articular space widening Condyle elongation and distortion	Yes	No	24
15. Sato et al (1977)	-	-	36	Pain	-	Yes	Yes	Not reported
15. Sato et al (1977)	-	-	18	Swelling, pain, and trismus	Radiopaque mass with radiolucent areas on TMJ	Yes	No	Not reported
15. Sato et al (1977)	-	-	4	Swelling, pain, and trismus	Radiopaque mass with radiolucent areas on TMJ	Yes	No	Not reported
16. Cadenat et al (1979)	F	60	-	Swelling and pain	-	Yes	No	6
17. Morris et al (1987)	F	29	24	Headache and swelling	Mass from condyle to infratemporal fossa; Cranial fossa eroded	Yes	Yes	6
18. Wasenko and Rosenbloom (1990)	F	49	-	Swelling and pain	Mass from condyle to infratemporal fossa with calcification	Yes	No	Not reported
19. Nitzan et al (1993)	F	36	72	Spontaneous pain and swelling	TMJ space radiolucent lesion: condyle resorption	Yes	No	84
20. Giraud et al (1997)	F	28	36	Mouth opening difficulty and swelling	Heterogeneous mass in temporal tubercles	Yes	Yes	27 no recurrence
21. Ichikawa et al (1998)	F	66	12	Trismus and pain	Mass surrounding the joint; Erosion of the condyle	Yes	No	36
22. Batra et al (1999)	M	65	18	Swelling and hearing loss	Mass just anterior to the left external ear canal and encasing the mandibular condyle	Yes	No	7
23. Angiero et al (2007)	F	64	7	Swelling and pain	Radiopaque mass extending from the skull base to the medial pterygoid space	Yes	No	96 No recurrence

*According to a Pubmed search using specific indexing terms.
†TMJ signs and symptoms.

Chondrosarcoma is the second most common primary spindle cell tumor of bone, but it is uncommon in the head or neck region.[5] It has been suggested that between 6.4% and 12% of all chondrosarcomas are located in the head and neck areas,[5–7] where the most common sites are the larynx, followed by mandible, maxilla, and maxillofacial skeleton.[8] Chondrosarcoma involving synovial tissues is even more rare. Merril et al[9] showed that the common sites for its occurrence are the knee, followed by the hip, ankle, groin, and elbow. Temporomandibular joint (TMJ) chondrosarcoma was described by Sesenna et al[3] as an exceptional occurrence. The age range of when this neoplasm occurs is from 17 to 75 years and the majority of these patients are in the third to fourth decades of life. There is no gender preference.[3,8,10–23]

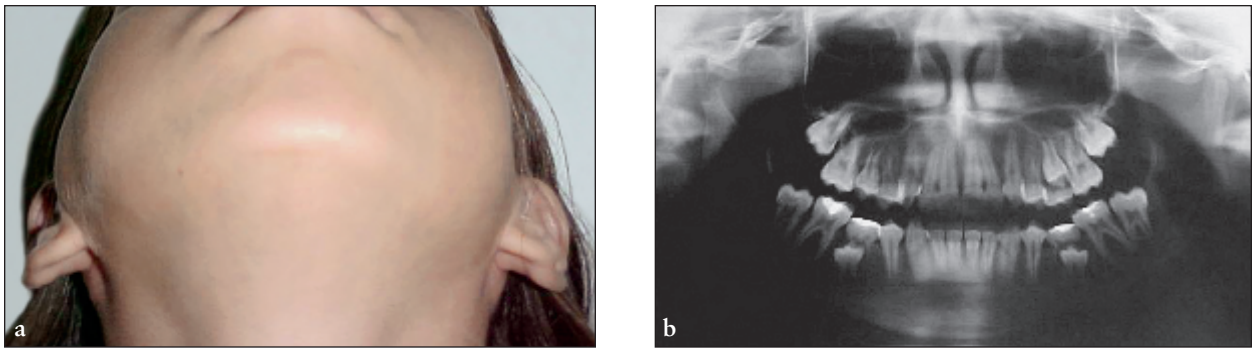


Fig 1 *(a)* Clinical aspect of facial asymmetry on the right preauricular area; *(b)* Panoramic radiograph showing the right condylar erosion compared to the left normal joint.

While tumors and tumorous masses arising as primary lesions of the TMJ are rare, the presentation of these lesions usually mimics that of much more common disorders, often leading to a delay in diagnosis. For example, patients often have unilateral facial swelling, external auditory canal obstruction, or chronic TMJ dysfunction at presentation.[8] Chondrosarcoma has no pathognomonic findings and cannot be differentiated from other conditions based on clinical signs.[4] TMJ chronic disorders, synovial chondromatosis,[9,21,24–26] chondroma,[3] benign chondroblastoma,[9] chondromyxoid fibroma,[9] synovial sarcoma,[23] and Ewing sarcoma[23] must be included in a differential diagnosis.

Radiographically, chondrosarcoma is associated with single or multiple radiolucent areas seen on plain films.[22] The most characteristic feature is a combination of bony destruction, often with intralesional calcifications, and a periosteal reaction. The intralesional calcifications are best seen on computer tomography (CT) scans. Magnetic resonance imaging (MRI) features vary according to the grade of the lesions.[27] Microscopically, these lesions appear as a lobulated cellular neoplasm composed of atypical, pleomorphic chondroblastic cells arranged in a hyaline matrix with occasional foci of calcification.[22] Studies have been carried out to try to develop criteria to distinguish synovial chondromatosis from chondrosarcoma.[24]

A Pubmed search of the English literature with specific indexing terms (chondrosarcoma, malignant neoplasm, synovial, and temporomandibular joint) yielded 20 TMJ chondrosarcoma cases published up to 2008 (Table 1). An additional case of chondrosarcoma of the TMJ is presented here. This appears to be the first TMJ chondrosarcoma in a child reported up until the present.

Case Report

An 11-year-old girl was referred to Centro Goiano de Doenças da Boca, School of Dentistry, Federal University of Goiás, Brazil, with the chief complaint of a right preauricular swelling (Fig 1). Her father had noticed it by chance about 2 months earlier. There was no history of trauma or any other event that contributed to the onset of the symptoms presented: discrete pain and mild limitation of mouth opening. There was no hearing loss and an aspiration biopsy was negative. The patient was in good health. On physical examination, she reported mild pain on palpation of the right preauricular region and the lesion presented as a tender mass.

A panoramic radiograph (Fig 1) revealed degenerative changes/erosion of the right condyle that had not been noticed in a similar x-ray examination one year earlier, which was requested by her orthodontist. A CT scan showed an osteolytic lesion in the right condyle. The low-density lesion extended both internally and externally to the masticator space, presenting discrete peripheral enhancement after contrast media injection. Enlargement of the ipsilateral mandibular fossa could be observed. The skull base, parotid, and carotid spaces were preserved (Fig 2). An ultrasound evaluation confirmed the solid content of the 27 × 19 mm lesion.

Spin Echo T1-weighted (SE T1W) MRI confirmed an invasion of the masticator space by the TMJ mass that exhibited an isointense signal relative to the muscles. The masticator space is one of the normal anatomical spaces of the suprahyoid neck, defined by the deep cervical fascia, and an MRI contrast description of the mass provides etiological hypothesis. The mass has a similar contrast to the water-based tissues, eg, muscle, cartilage.

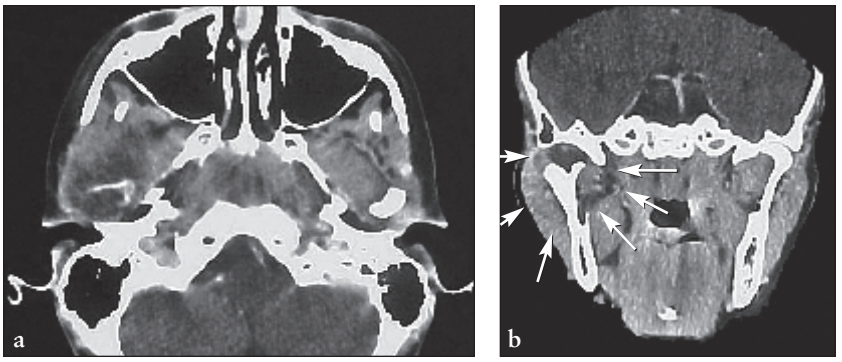


Fig 2 *(a)* Axial and *(b)* coronal CT on soft tissue window revealed the osteolytic lesion on the right condyle invading the surrounding anatomic structures *(arrows)*.

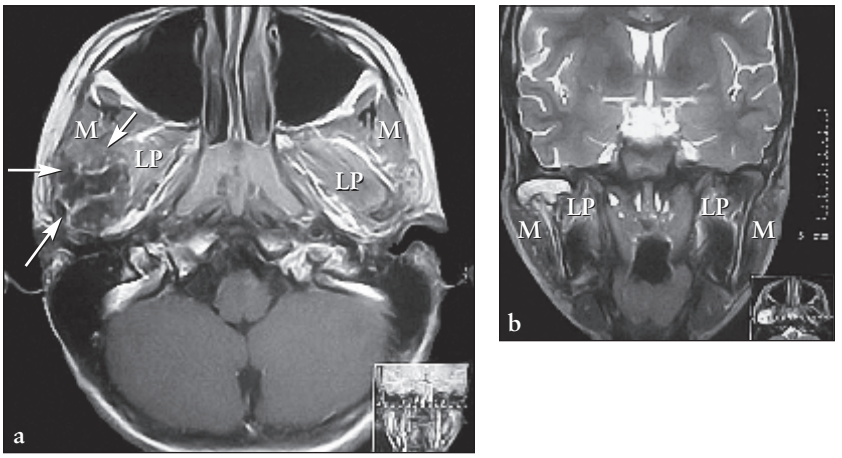


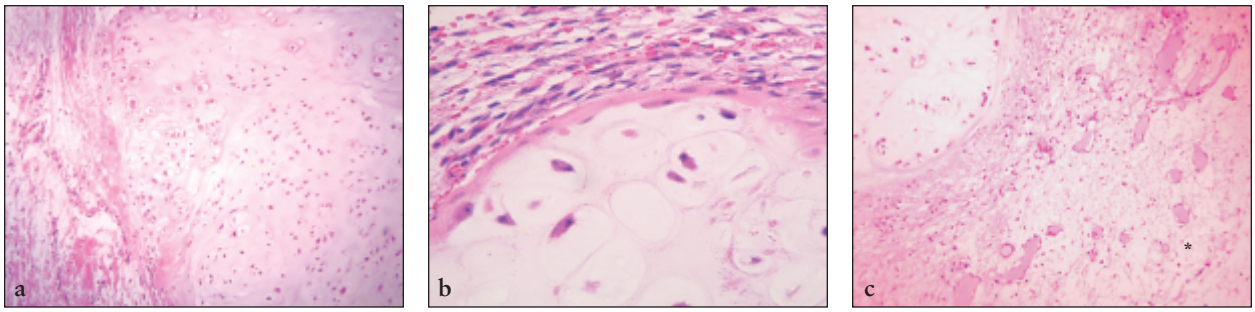
Fig 3 SE MRI *(a)* Axial T1W postcontrast image shows a mass *(arrows)* with a heterogeneous postcontrast enhancement in right TMJ and masticator space topography. This cervicofascial space encloses muscles as lateral and medial pterygoid, masseter and temporalis; inferior alveolar nerve, ramus, and body of mandible. *(b)* Coronal T2W image reveals the "filling-up" pattern of tumor growing into the right condyle. (M = masseter muscle, LP = lateral pterygoid muscle, inserts = M and LP muscles).

Lateral displacement of the masseter muscle and medial displacement of the lateral pterygoid muscle could be observed. A heterogeneous contrast enhancement was observed after paramagnetic contrast injection (Fig 3). On T2-weighted (T2W) images, the lesion also had a heterogeneous hyperintense signal. Permeation of trabecular bone with "filling-up" of condyle marrow spaces could also be seen on SE T2W images (Figs 3 and 4).

After an incisional biopsy of a representative portion of the lesion, the specimens were fixed in 10% neutral buffered formalin, embedded in paraffin, and 4-μm sections cut and stained with hematoxylin and eosin. Serial sections were evaluated immunohistochemically for Ki-67 (clone MM1, Novocastra; 1:100), Cyclin B1 (clone 7A9, Novocastra; 1:40), and p53 (clone DO-7, Novocastra; 1:200) proteins, using the labeled streptavidin-biotin-immunoperoxidase detection system (LSAB kit; K0492, DAKO). Appropriate positive controls were prepared for each antibody while negative controls consisted of slides that had not been treated with the primary antibodies.

The microscopic examination showed a well-differentiated cartilaginous tissue proliferation with some cellular pleomorphism and hyperchromatic nuclei, permeated by fibrous stroma (Fig 4). In addition, rare mitotic figures and a low proliferative index, measured by staining neoplastic cells with the Ki-67 and Cyclin B1 antibodies, were evident. Only a few p53 positive neoplastic cells were observed. After these microscopic features were considered, a diagnosis of low-grade chondrosarcoma was established. A high condylectomy was performed. The diagnosis from the incisional biopsy was confirmed and margins of the surgical specimen were tumor free.

One year after surgery, the patient presented mild pain in the temporalis muscle during mastication which was controlled by physical therapy. Three and a half years follow-up (Fig 5) did not show any metastasis or recurrence. Hyperplasic lymph nodes on the right cervical region were identified during palpation. An ultrasound examination showed an undetermined vascularization pattern of the bilateral lymph nodes, reinforcing the need for fine needle biopsy cytology which suggested a



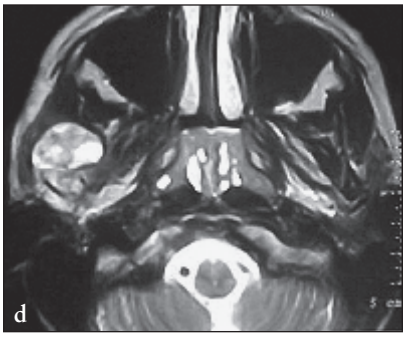


Fig 4 *(a)* Low-powered photomicrograph of the right condyle shows a well-differentiated cartilaginous tissue permeated by fibrous stroma. *(b)* In detail, areas of spindling at the periphery of the cartilage with hypercellularity and nuclear pleomorphism. (Hematoxylin-eosin, original magnification ×100 *[a]* and ×400 *[b]*). *(c)* Mixoid (*) and cartilaginous areas of the neoplasm (Hematoxylin-eosin, original magnification ×100) are probably related to the heterogeneous hyperintense signal of the mass seen on *(d)* T2W image (Axial T2W image). This is not a pathognomonic chondrosarcoma image pattern, but it provides additional information for differential diagnosis, reinforcing the hypothesis of a chondromyxoid lesion.

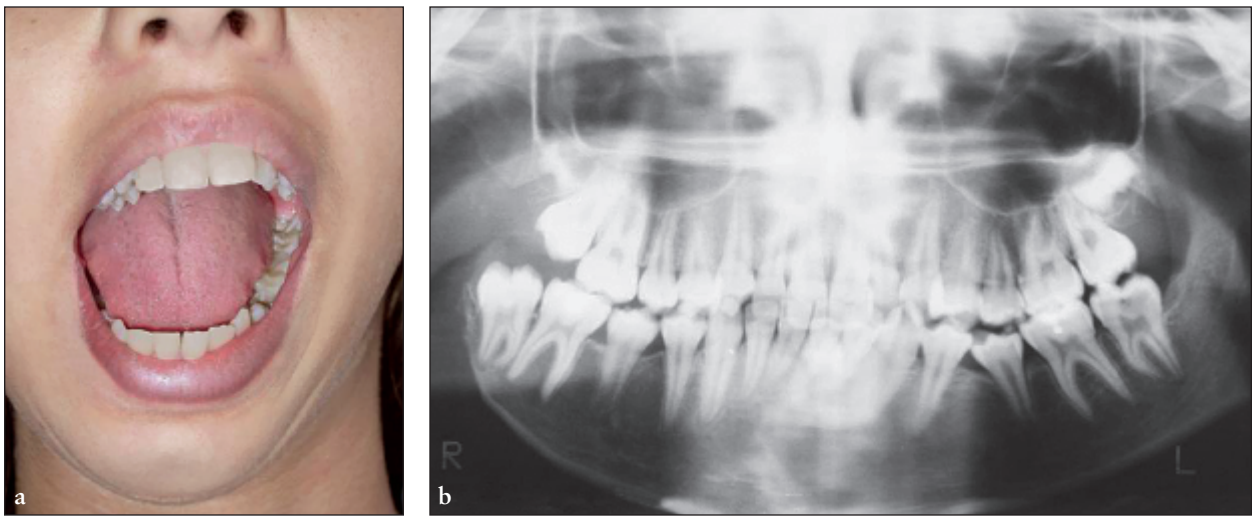


Fig 5 *(a)* Clinical and *(b)* radiographic aspects of the patient at 3.5 years follow-up.

reactive process, and only a follow-up was recommended. The patient still complained about the temporalis pain, which she was able to control with analgesic drugs. An occlusal splint and/or laser therapy of the muscle, as well as continuing physical therapy, were recommended and she has been evaluated regularly. Six years after surgery, the patient remained without metastasis and underwent orthodontic treatment, after which the muscle pain became sporadic. The patient also received authorization from the surgeon/oncologist for oral rehabilitation (graft/implant).

Discussion

From 1954 to 2008, 20 cases of TMJ chondrosarcoma were published in the English literature and the youngest patient was 17 years old. All the others were between the second and seventh decades of life (Table 1). This reinforces how rare a chondrosarcoma is in an 11-year-old child, making the present case the first ever reported for this age. Table 1 shows that swelling[3,8,10–23] and pain[10–12,15,16,18,19,21–23] have been cited as the chief complaints. The patient in the present case

presented discrete pain and preauricular swelling. All cases reported in the reviewed literature were submitted to surgical treatment and only four went through irradiation treatment as coadjuvant therapy. The present case did not have irradiation as part of treatment.

TMJ chondrosarcoma presents nonspecific symptoms and patients usually complain of swelling in the preauricular region and discrete pain.[28] This tumor grows slowly and can be asymptomatic until it reaches a considerable size. The duration of symptoms has been reported to be quite variable, ranging from several weeks to more than one year.[5] In the present case, the neoplasm was identified by chance by the patient's father, who believed that the lesion had been developing for 2 months at the first consultation. It took another 2 months for that patient to complete all the complementary examinations (eg, CT, MRI, biopsy, histopathological), some of which were high-cost. The final histopathological diagnosis was widely discussed and it was established 4 months after the probable onset of injury.

There are no imaging pathognomonic findings associated with chondrosarcoma. In the reported case, CT demonstrated local bone destruction involving the condyle and an expansive mass surrounding the joint. MRI in this case was important for a differential diagnosis, as well as for the detailed information on the anatomic limits of the lesion for preoperative planning. The lesion isointense signal to the muscle on the T1WSE image supported the hypothesis of its hypercellular content. The heterogeneous hyperintensity on T2WSE image suggested the presence of a chondroid or myxoid matrix (Fig 4d).

Chondrosarcomas have been considered one of the most difficult malignant bone tumors to be distinguished from the standpoint of histopathology.[29] They were initially histologically graded by Evans et al[2] considering the cellularity of the lesion and the nuclear changes of the chondrocytes. In this three-grade system of differentiation, grade I tumors are well-differentiated with a growth resembling a benign cartilage tumor, whereas grade III lesions are aggressive and poorly differentiated with an increased number of cells with binucleate forms and mitotic figures. The more the neoplasm is undifferentiated, the more aggressive it is locally and the higher its potential to metastasize.[30]

A chondrosarcoma in joints shows clinical, radiographic, and histopathological features similar to those of synovial chondromatosis.[9,21,24–26] Primary synovial chondromatosis should be considered in the differential diagnosis of the monarticular presentation of an intra-articular soft tissue mass, particularly in the presence of bone erosions.[26] Permeation of trabecular bone with "filling up" of marrow spaces should be considered a sign of malignancy. According to Bertoni et al,[24] the most useful features for diagnosis are the following: *(1)* loss of the typical clustering pattern with abundant matrix juxtaposed to areas where the tumor cells are arranged in sheets; *(2)* myxoid change in the matrix; *(3)* hypercellularity with crowding and spindling of the nuclei at the periphery of the lesion; *(4)* presence of necrosis; *(5)* permeation of the trabecular bone and invasion of marrow spaces. The present case showed at least three of these criteria: myxoid change, areas of spindling at the periphery, and the "filling-up" pattern of the neoplasm growing into the condyle (Figs 3b, 4b, 4c, and 4d). Therefore, the histopathological content suggested by MRI was confirmed by the anatomopathologic findings (Fig 4).

The well-differentiated aspect of the specimen resulted in a final diagnosis of synovial chondrosarcoma of low malignancy grade. Although a high expression of the cellular markers of biological activity, such as Ki-67 and p53, may indicate greater aggressiveness in the tumor[31,32] and their evaluation has been suggested to distinguish low- from high-grade chondrosarcomas,[27,31,32] this is not yet considered of great clinical relevance.

The radiographic appearance of the tumor, its hypercellularity with crowding and spindling of the nuclei at the periphery of the lesion, and the presence of atypical condrocytes were useful in distinguishing this tumor from chondroma,[28] benign chondroblastoma,[9] and chondromyxoid fibroma,[9] all three benign neoplasms, and from chondroblastic-osteosarcoma, which was excluded because of the abscence of a malignant osteoid matrix.[28]

Wide local resection is the treatment of choice for patients with chondrosarcoma of the TMJ. Although it is traditionally regarded as a radioresistant tumor,[33] some authors have recommended radiation as a postoperative adjuvant therapy for patients with high-grade tumors.[7,34] In the present case, the patient was only subjected to surgical treatment.

The clinical behavior of this tumor is variable and linked to histologic grading.[8] Local recurrence is more common than distant metastasis and is more dependent on the adequacy of surgical resection than histologic grade.[2] But recurrence can occur after 5 or 10 to 20 years.[34] Chondrosarcoma of the jaws has a poor prognosis. After several years without recurrence, the present case is still under periodic clinical evaluation.

In conclusion, cartilaginous lesions in the jaws must always be regarded with extreme suspicion since benign and malignant lesions show similar clinical features but a different prognosis and treatment, and a misdiagnosis can compromise patient survival rate. This case emphasizes the importance of interdisciplinary approaches to minimize the possibility of misdiagnosis.

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