Chondroid Tenosynovial Giant Cell Tumor of Temporomandibular Joint

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Abstract

Tenosynovial giant cell tumor (TGCT), a benign proliferative lesion arising from the synovial membrane of the joints, is rarely seen in the temporomandibular joint. It frequently presents as a painful, preauricular swelling and affects the jaw functions. Two types of TGCT seen are diffuse TGCT (TGCT-D) and localized TGCT. A case of TGCT-D is described here, the highlight of the case being extensive areas of chondroid metaplasia which mimicked chondroid lesions, but was ruled out based on negative immunohistochemical findings within the tumor cells.

Keywords: Chondroid metaplasia, synovial membrane, temporomandibular joint, tenosynovial giant cell tumor

Introduction

The pathological swellings in the temporomandibular joint (TMJ) can be due to various tumors and pseudotumors such as osteochondroma, chondrosarcoma, osteoid osteoma, plasmacytoma, synovial chondromatosis, eosinophilic granuloma, and pigmented villonodular synovitis.[1] Most of these tumors are rare and hence often clinically diagnosed as parotid mass or infection.[2] One example is tenosynovial giant cell tumor (TGCT), which is a rare benign proliferative growth of the synovium which shows destructive invasion by synovial-like mononuclear cells.[3]

TGCT, being one of the most common soft-tissue tumors of the hand (finger joints and tendon sheaths), is rarely seen in the head or neck region.[4,5] According to the WHO classification of 2002 and 2013, the incidence rate in TGCT is not exactly known.[6] A total of 73 cases of TGCT affecting the TMJ have been reported in the English literature.[7] They are divided into four clinicopathological subtypes, according to their growth pattern (localized or diffuse) and anatomical site (intra- or extra-articular).[8] Difference between localized TGCT (TGCT-L) and diffuse TGCT (TGCT-D) is that TGCT-D has a tendency to occur in younger individuals, presents as a less well-defined soft-tissue mass, and has a locally aggressive growth and a high recurrence rate.[4,5] This report presents a case of TGCT-D in the TMJ with chondroid metaplasia.

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Case Report

A 26-year-old male patient reported with the complaint of pain and swelling in the right pretragal region in 2015. The patient had a history of swelling and pain in the same region 5 years back, for which he had undergone curettage of the right condyle and superficial parotidectomy at a government medical institute. Histopathology report then showed normal salivary gland tissue and benign giant cell tumor of the condyle. The patient was asymptomatic until 4 months back when he noticed similar swelling associated with pain. On examination, a healed surgical scar was noted on the right preauricular region and extraoral diffuse swelling of about 4 cm in diameter in the right pretragal region [Figure 1a and b].

Computed tomography (CT) showed a large expansile hypodense lytic lesion with lobulated margins measuring 2.7 cm × 2.9 cm × 2.6 cm involving the articular surface of the condylar process with thin bony septa within it [Figure 2].

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Figure 1: Frontal view of the clinical photograph showing extraoral diffuse swelling of about 4 cm in diameter in the right pretragal region (a). Lateral view of the lesion with healed surgical scar noted on the right preauricular region (b)

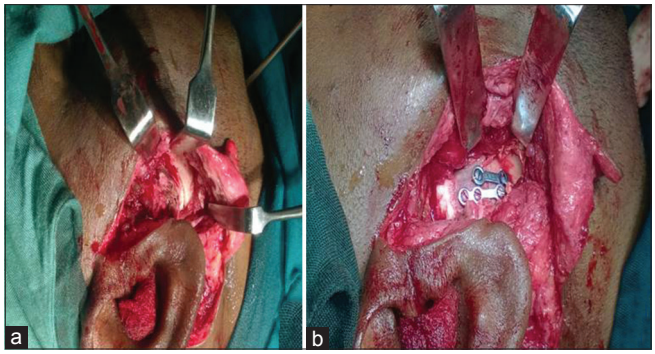

Figure 3: Intraoperative photograph showing grossly deformed condyle (a) and surgical resection of the right condyle and coronoid with reconstruction using costochondral rib graft (b)

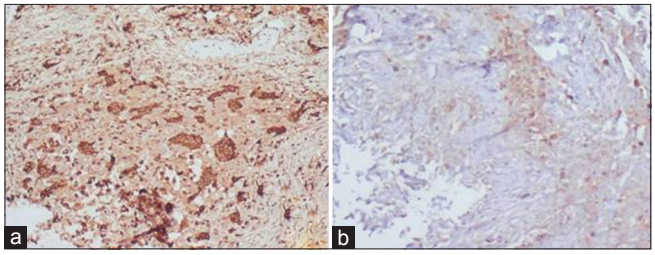

Figure 5: The epitheloid cells show positive staining with CD68 (a) and negative with S-100 (b) (×100)

Surgically, on inspection, the condyle was grossly deformed and reddish brown in color. Further anterior dissection was done till the coronoid process and associated part of superior ramus. Surgical resection of the right condyle and coronoid was done and reconstructed using costochondral rib graft [Figure 3a and b].

The histopathological examination of the biopsied tissue revealed a poorly circumscribed hypercellular connective tissue stroma [Figure 4a]. Stroma showed areas of hyalinization, consisting of lot of plump spindle-shaped cells and epitheloid mononuclear cells. It also showed extensive areas of hemosiderin pigmentation and lot of multinucleated giant cells [Figure 4b]. Diffuse

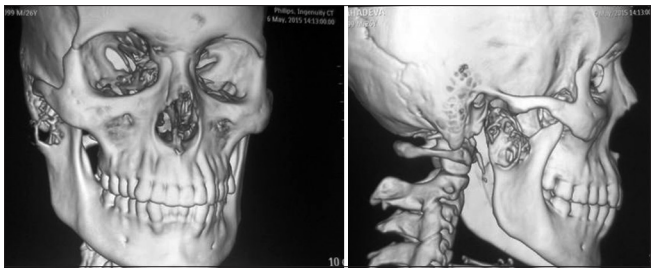

Figure 2: Computed tomography scan showing a large expansile hypodense lytic lesion with lobulated margins measuring involving the articular surface of the condylar process

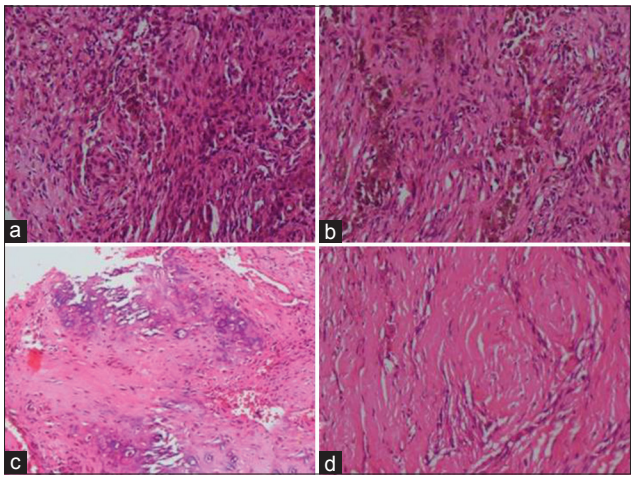

Figure 4: Sections reveal a poorly circumscribed hypercellular connective tissue stroma (a). Stroma showed areas of hyalinization, with lot of plump spindle-shaped cells, epitheloid mononuclear cells, extensive areas of hemosiderin pigmentation, and lot of multinucleated giant cells (b). Diffuse areas of chondroid metaplasia (c) and few hypocellular connective tissue areas were also evident (d) (H and E, ×100)

areas of chondroid metaplasia were also evident within the stroma [Figure 4c]. Few hypocellular connective tissue areas were also evident [Figure 4d]. Based on the above findings, a diagnosis of chondroid TGCT was given. Immunohistochemistry (IHC) with CD68 was positive [Figure 5a] and negative with S-100 [Figure 5b] in the epitheloid mononucleated giant cells, further confirming the diagnosis of TGCT and ruling out chondroid lesions such as chondroblastoma and chondrosarcoma. The case has been followed up regularly, and the 2-year follow-up has been uneventful.

Discussion

TGCT belongs to a group of proliferative lesions affecting the synovial membrane and the tendon sheath, and it is rarely seen in the head or neck region. The etiology of TGCT is not certain. However, recent advances indicate that TGCT is a clonal neoplasm driven by overexpression of colony-stimulating factor-1.[2] TGCT-D differs in its clinical presentation from TGCT-L as it is more common in young people and presents as a poorly circumscribed mass with local aggressive biological behavior and high recurrence rates. Another distinct feature

from TGCT-L is that it does not have a fibrous capsule.[4] The present case was a locally aggressive, recurrent growth in a young individual and did not show any fibrous capsule.

Radiologically, TGCT shows a soft-tissue mass often causing bone erosion and scalloped margins. CT scan frequently reveals cyst formation and areas of hyperattenuation due to hemosiderin deposition.[9] CT scan in our case revealed a large hypodense lytic lesion.

Histopathologically, TGCT shows features of synovial hyperplasia, with polyhedral synovial histiocyte-like cells, along with varying proportion of lymphocytes, giant cells, macrophages, and hemosiderin pigmentation.[3] A variant of TGCT called the "chondroid TGCT" in addition shows chondromyxoid, chondro-osseous, and hyaline-like matrix patterns.[10] The histopathology in the present case was suggestive of TGCT, with hyaline-like and chondroid areas. In view of the presence of large chondroid areas in the present case, IHC was done with CD68 and S-100. In TGCT, the histiocytes and multinucleated giant cells stain positively with CD68.[11] S-100 positivity in these cells is used to distinguish chondroid tumors which show positive staining.[12] Other markers that have shown varying positive expression are clusterin, desmin, CD163, CD68, and S-100. S-100-positive nuclei were found in the metaplastic chondrocytes in cartilaginous metaplasia, while the surrounding synovium-like monocytes, small histiocytoid cells, and multinucleated giant cells were S-100 negative.[7] In our case, the cells were CD68 positive and S-100 negative, thus ruling out the possibility of these cells being chondrocytic. Therefore, a final diagnosis of TGCT-D type with chondroid metaplasia was given based on the clinical, histopathological, and IHC findings.

Although surgical excision is considered as a preferred method of treatment for all forms of TGCT, it is influenced by the nature of the lesion. In most of the cases described in the literature, the treatment was with selective surgery, including enucleation of the neoplasm with wide surgical margins.[11] In the current case, since there was extensive recurrent involvement, resection of involved part was done and reconstructed to reduce the morbidity. During 2-year follow-up, there has been no evidence of recurrence.

Conclusion

TGCT is a benign tumor which can be locally aggressive and a concern for high morbidity. Its occurrence in TMJ is rare, but establishing correct diagnosis aids in proper treatment planning and prognosis to the patient. In cases, where other features such as predominant chondroid areas cause difficulty in arriving at its definitive diagnosis, molecular investigations such as IHC with specific markers are advised.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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