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Protuberant fibro-osseous lesions of the temporal bone a case report **BRIJESH KUMAR TIWARI**

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Abstract : Ossifying fibroma (OF) of the temporal bone is a rare entity, and constitutes a minor proportion of fibro-osseous lesions of the head and neck. Although these tumors are primarily described in the maxilla and mandible, a few cases have been reported describing that these tumors originate from paranasal sinuses and orbit. Here we report a case of a recurring ossifying fibroma of the temporal bone and discuss the management of these tumors, highlighting their tendency to recur despite complete excision.

Keyword :Ossifying fibroma, Temporal bone, Overlaping histological feature.

Introduction

Ossifying fibroma (OF) as a distinct entity was first described by Menzel in 1872, although the term 'ossifying fibroma' was coined by Montgomery in 1927.(1) It is a benign fibro-osseous lesion and typically occurs in the maxilla or mandible; however, there have been isolated reports of temporal bone involvement in literature. (2)(3)(4)These tumors are characterized by slow growth and tend to produce symptoms due to compression of adjacent structures, however they tend to grow aggressively in younger patients. Treatment for these tumors is surgical excision. Early aggressive surgery has been advocated as they tend to recur, despite being totally excised on most occasions. We hereby describe a case of ossifying fibroma of the temporal bone with overlapping histological features of giant cell lesion, which recurred despite total excision.

Case report

An eighteen-year-old male presented to our outpatient with a history of a mass protruding from his left ear associated with progressive hearing loss. On examination he had a conductive hearing loss in his left ear and there was a polypoidal mass measuring 4 x 4 x 3 cms, protruding out of the external auditory canal. There was no discharge from the swelling. He had no focal neurological deficits. Magnetic resonance imaging (MRI) and computed tomography (CT) showed a large soft tissue mass completely filling the left external auditory canal with extension into the middle ear cavity and the mastoid antrum with multiple fluid-fluid levels. There was no intradural extension of the tumor. (Fig 1,2.)

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Fig 2: CT (bone window) of the temporal bone showing a osteolytic mass, eroding the left petrous bone and filling the left external auditory canal.

We considered a differential diagnoses of a giant cell tumor with secondary aneurysmal bone cyst (ABC) formation, telangiectatic osteosarcoma or a solitary fibrous tumor. He underwent a left temporal craniotomy and radical excision of the mass, along with a left radical mastoidectomy and culdesac closure of the left external ear performed by the otolaryngologists. Histopathology was reported as fibrocollagenous tissue lined by keratinizing stratified squamous epithelium with focal surface ulceration covered by acute inflammatory exudate.

The subepithelial stroma was infiltrated by a tumor composed of interlacing fascicles and sheets of polygonal to spindle

shaped cells with moderately pleomorphic vesicular nuclei, prominent eosinophilic nucleoli and moderate amounts of eosinophilic cytoplasm and indistinct cell borders. Mitotic activity was inconspicuous. Numerous osteoclast like giant cells were seen scattered throughout the lesion. Also seen closely associated with tumor cells were several scattered curvilinear trabeculae of osteoid and woven bone. There were moderately dense perivascular and subepithelial infiltrates of inflammatory cells composed of lymphocytes, plasma cells, histiocytes and few hemosiderin-laden macrophages. Several cystic spaces some of which contained blood and lined by osteoclastic giant cells were present. There was no evidence of malignancy. These histopathological features were suggestive of an ossifying fibroma. There were overlapping features of a giant cell rich benign fibro-osseous lesion with secondary aneurysmal bone cyst formation. (Fig 3-7) He subsequently had an uneventful postoperative recovery.



Fig 3 (A,B): Curvilinear trabeculae of reactive new bone and few seams of osteoid along with scattered osteoclast-like giant cells.



Fig 4: Low power view of osteoclast like giant cells.



Fig 5: Fascicles and sheets of polygonal to spindle shaped tumour cells are seen exhibiting moderately pleomorphic vesicular nuclei, conspicuous eosinophilic nucleoli and moderate amounts of eosinophilic cytoplasm with indistinct cell borders (H&E 40X).



Fig 6: Cystic space containing blood and lined by osteoclastic giant cells.



Fig 7: Focal xanthomatous reaction with moderate infiltrates of chronic inflammatory cells and focal cholesterol clefts.

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He presented to us 10 months later for a routine follow up, with no new symptoms. A repeat MRI brain showed recurrence of the tumor in the left temporal bone measuring $4.2 \times 3.6 \times 3.4$ cm. It was expansile, multiloculated, with blood fluid levels, and showed peripheral contrast enhancement. It extended into the middle cranial fossa, causing significant compression of the left temporal lobe with effacement of the temporal horn. However there was no obvious intradural extension. (Fig 8.)



Fig 8(A,B): Follow up MRI brain showing recurrence of the tumor and significant compression of the left temporal lobe. (A) T2 weighted axial image and (B) T1 weighted axial image with gadolinium injection.

He underwent re-exploration and radical excision of the recurrent tumor, which was found to have grown into the previous mastoidectomy defect and also extended into the inner ear. The histopathology was reported to be similar to the previous one. Postoperatively, he had an uneventful recovery and a postoperative CT brain showed a small cystic collection in the left middle cranial fossa with no obvious residual tumor. (Fig 9.) However, due to recurrence of this tumor despite a radical excision, we advised the patient to undergo conformal radiotherapy.



Fig 9: Postoperative contrast CT brain showing total excision of the tumour with a small cystic collection in the left middle cranial fossa. Discussion

Protuberant Ossifying fibroma of the temporal bone has been reported in literature, but whether they are a distinct entity or they belong to a spectrum of benign fibro-osseous lesions, still remains unclear. A fibroma of the temporal bone was first reported by Stecker in 1971. (5)Another similar entity described in literature has been named protuberant fibro-osseous lesion. It is also occasionally referred to as 'Bullough lesions'. (6) These tumors in the temporal bone, are benign, but locally invasive, and produce symptoms mainly by invasion and compression of adjacent structures. Patients commonly present with headache, tinnitus, aural fullness, purulent otorrhea or in the later stages, with a protruding mass from the external auditory canal. Very rarely do these tumors present with features of intradural extension into the adjacent temporal lobe. Radiologically, ossifying fibromas are well circumscribed with a unilocular or multilocular appearance, and varying degrees of mineralizationm within them. The differential diagnoses to be borne in mind for such cases include the following: osteomas or exostoses, osteoblastoma, invasive or ectopic meningiomas, giant-cell granulomas, aneurysmal bone cysts, eosinophilic granulomas, and especially fibrous dysplasia. (7)(8)(5)There is a considerable radiological and pathological overlap between Ossifying fibroma,

fibrous dysplasia and also giant cell tumors with secondary Aneurysmal bone cyst formation, as was demonstrated clearly in our case. Radiologically, the multiple fluid-fluid levels resembled those seen in a classical ABC, and pathologically it was predominantly a fibro-osseous lesion, with a giant cell rich component and a secondary aneurysmal bone cyst formation. The pathogenesis of these tumors is not clearly known, while it has been speculated that OF maybe a reactive lesion, secondary to a trauma suffered in the past. (2) Although well circumscribed, these tumors bear the potential of pathological disruption of the proximate structures of the temporal bone. Despite being benign, their locally aggressive nature warrants an early and complete surgical excision of these tumors, to prevent recurrence. However, even a complete excision does not necessarily eliminate the chances of recurrence, as is evident from our case. Surgical excision remains the gold standard of treatment, however the role of adjuvant treatment (radiotherapy or chemotherapy) still remains unclear. (4)(3) Due to the rarity of these tumors, prospective studies comparing various modalities of treatment have not been performed. In our case, the tumor was radically excised, only to recur within a period of 10 months. Hence close follow-up is mandatory for these tumors, and if needed re-excision remains the only option of treatment in recurrent cases. Conclusion

Ossifying fibroma of the temporal bone is a rare entity, which is benign and locally aggressive. It requires a detailed histopathological examination to distinguish it from its mimicks. At present, early and radical surgical excision remains the treatment of choice. However, it is recommended to keep these patients under close follow-up, as these tumors notoriously tend to recur.

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