Temporal Bone an Unusual Site for Juvenile Psammomatoid Ossifying Fibroma

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Short History

A 14-year female presented with a complaint of painless gradually increasing swelling in the right temporal region for 4 years. However, the patient complained of pain from the swelling for 1 week before admission. On examination, a smooth surfaced slightly tender bony lesion measuring approximately 8x7.5cm was noted in the frontotemporal region. No neurological deficit was noted. High-resolution Computed Tomography (HRCT) showed a multiloculated expansile lytic lesion of the temporal bone. The lesion involves the squamous part of the right temporal bone showing thin internal septations and a peripheral sclerotic rim along with the extra-axial intracranial extension. Contrast-enhanced magnetic resonance tomography (CEMRI) showed a multiseptated lesion in the right squamous temporal bone with characteristic signal intensities with a T2 hypointense rim (Fig 1). The patient underwent surgical excision of the neoplasm. Gross examination showed a bony tissue bit weighing 65 grams and measuring (9x7.5x4) cm. The outer surface was bony and hard in consistency inner surface showed multiple multiloculated cystic blood-filled spaces. Light microscopic examination showed cellular fibroblastic stroma consisting of spindled cells without any nuclear atypia. Embedded in the fibroblastic stroma were multiple basophilic small-sized ossicles (psammomatoid bodies). Also noted are fibrous septa separated cystic spaces filled with blood. The fibrous septa are composed of fibroblasts, multinucleated osteoclastic type giant cells, and osteoblast-rimmed woven reactive bone. No atypical mitosis areas of necrosis or cellular atypia were noted (Fig 2). Epithelial Membrane Antigen (EMA) was negative in the tumor cells. Based on the clinical-radiological-histopathological findings a diagnosis of Juvenile psammomatoid ossifying fibroma (JPOF) with an aneurysmal bony cyst-like area was given. The patient was reviewed 2 months post-surgery and no recurrence or any neurological deficits were noted.

Differential Diagnosis:

A wide range of differentials may be considered clinic-radio-pathologically in a case of JPOF. Fibrous dysplasia, osseous dysplasia, cemento-osseous dysplasia, osteoblastoma, osteosarcoma, Burkitt lymphoma and extra dural meningioma are some of the commonly considered differential diagnosis.[1]

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Discussion

As implicated by its name, JPOF a disease of the children and young adults shows no sex predilection.[2] Clinical findings in a case of JPOF ranges from being an asymptomatic bony-hard mass to a rapidly growing mass causing local destruction.[3] Radiological investigation usually shows well-demarcated uni or multiloculated radiopaque lesion without soft tissue involvement. The inner core of this lesion may be radiopaque-radiolucent or may show mixed opacities.Psammomatoid ossicle in JPOF presents as ground glass radiotransparent opacities and may mimic a cystic mass lesion.[4] Histologically juvenile trabecular ossifying fibroma (JTOF) and JPOF have identical fibroblastic stroma however in cases of JPOF multiple lamellate to spherical psammomatoid ossicles resembling meningiomatous psammomatoid bodies are characteristically noted. Incomplete or partial resections of JPOF are prone to recurrence hence a complete surgical resection is the preferred modality of treatment.[5] JPOF of the temporal bone is an extremely rare ossifying fibroma; the location is unusual for this neoplasm as it is primarily localized to the paranasal sinus and orbital bone, clinical findings aided by radiology and histopathology and occasionally immunohistochemistry findings are of utmost importance in the diagnosis of such neoplasms considering their propensity to recur.[6]

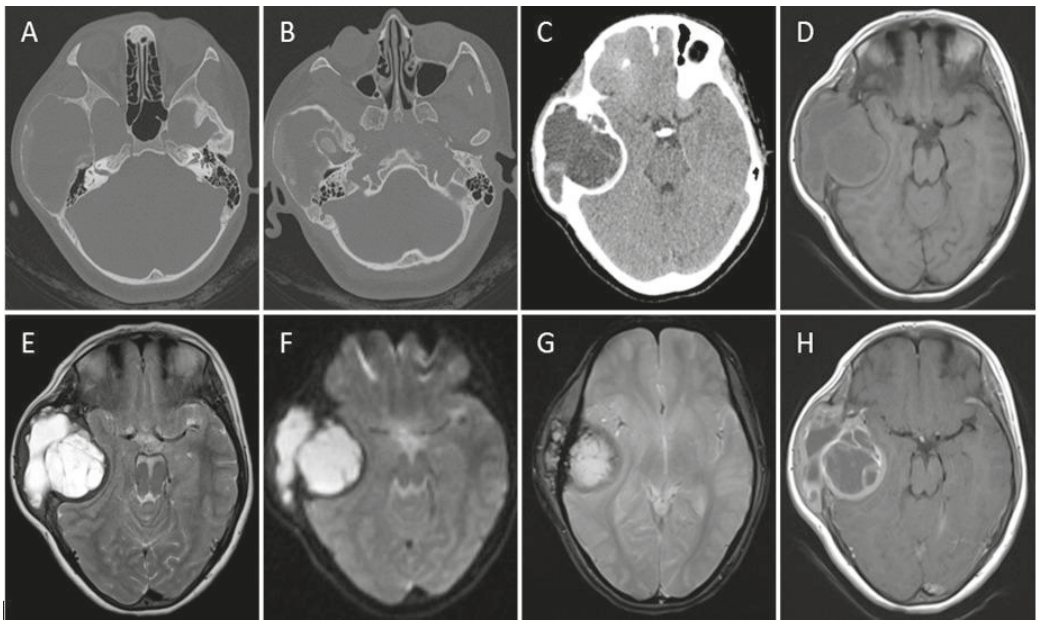

Fig. 1: (A, B) Axial sections of HRCT of temporal bone showing a multiloculated expansile lytic lesion centered in the squamous part of right temporal bone with peripheral rim of sclerosis and thin internal septations in the absence of cortical disruption **(C)** NCCT axial section in soft tissue window showing hypoattenuating contents within the lesion **(D)** T1-weighted and **(E)** T2-weighted MR axial images showing the lesion following fluid signal characteristics with internal septations and no evidence of fluid-fluid levels. Note the T2-hypointense rim. **(F)** Trace diffusion image showing marked restriction within the lesion (the corresponding ADC map, revealing low signal, is not shown here) **(G)** Gradient echosequence image demonstrating few intralesional foci of blooming **(H)** post-Gadolinium T1-weighted axial image showing avid homogenous enhancement of the septae and wall.

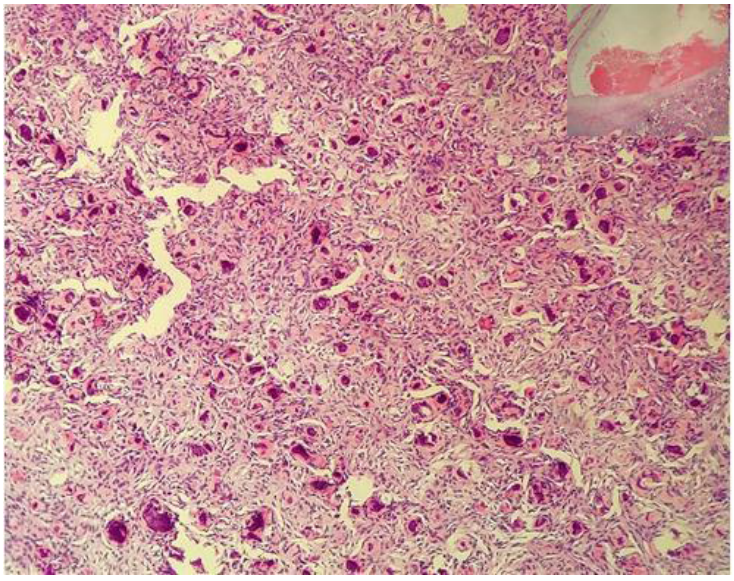

Fig. 2: Histomorphology showing psammomatoid bodies embedded in cellular fibroblastic stroma consisting of spindled cell without any nuclear atypia. (H and E, 200x). Inset showing aneurysmal bone cyst like areas. (H and E, 200x)

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