CASE REPORT

"Hybrid" lesion of the maxilla

Sankaranarayanan S, Srinivas S[1], Sivakumar P, Sudhakar R, (Capt.) Elangovan S[2]
Departments of Oral and Maxillofacial Pathology, [1]Orthodontics and Dentofacial Orthopaedics, Karpaga Vinayaga Institute of Dental Sciences, Chinna Kolambakkam, Kanchipuram Dt, [2]Department of Oral Medicine and Radiology, Ragas Dental College and Hospital, Uthandi, Chennai, India

Address for correspondence:
Dr. Sudhakar R,
Reader, Department of Oral and Maxillofacial Pathology, Karpaga Vinayaga Institute of Dental Sciences, Chinna Kolambakkam, Kanchipuram Dt, Tamil Nadu, India.
E-mail: dcprodent@gmail.com

ABSTRACT

Juvenile ossifying fibroma is an uncommon benign but aggressive fibroosseous lesion that affects the craniofacial skeleton. Their distinct clinical and histopathological features warrant the lesion to be considered as a separate entity from other fibro-osseous group of lesions such as fibrous dysplasia and cemento ossifying fibroma. Concomitant development of secondary aneurysmal bone cyst may rarely occur, which makes the lesion more aggressive and difficult to treat. We report a case of a 6 year old girl who was diagnosed with aneurysmal bone cyst during her earlier presentation at a private hospital and was treated for the same. The lesion recurred within 6 months. The second incisional biopsy specimen revealed features of trabecular variant of juvenile ossifying fibroma along with areas of aneurysmal bone cyst.

Key words: Hybrid lesion, aneurysmal bone cyst, juvenile ossifying fibroma, trabecular variant

INTRODUCTION

Juvenile ossifying fibromas (JOF) are rare fibro-osseous neoplasms that arise within the craniofacial bones in individuals under 15 years of age.[1] Clinicopathologically, JOF presents as two distinct forms, trabecular juvenile ossifying fibroma (TrJOF) and psammomatoid juvenile ossifying fibroma (PsJOF).[2]

Aneurysmal bone cysts (ABC) are osteolytic lesions containing blood filled spaces. They can present either as a primary lesion by itself or as a secondary change in a pre-existing lesion.[3] Cases of JOF with secondary ABC like areas, though uncommon, have been reported in earlier literature.[4-11] The importance of such presentation lies in the fact that cases of JOF associated with secondary ABC tend to show a more aggressive growth pattern and greater recurrence potential.[12]

CASE REPORT

A 6-year-old girl came to our institution with a swelling over the left side of the face which had been increasing in size since 4 months. She had consulted a private dental surgeon six months earlier with a similar complaint of progressive swelling in the same region. Clinical examination, CT imaging of the lesion, and incisional biopsy were performed. The histopathological report then was suggestive of an aneurysmal bone cyst and curettage of the lesion was performed under general anesthesia.

At present the patient had a diffuse swelling over the left middle third of face [Figure 1]. The skin over the swelling was firm and mildly tender. Intraorally, the left side of the hard palate had a soft, nontender swelling of around 3 × 3 cms in size [Figure 2]. The upper left buccal sulcus was obliterated from the canine to second molar and egg shell crackling could be felt on palpation. No decayed or nonvital teeth were present.

Aspiration through buccal aspect yielded blood tinged fluid. All routine hematological and biochemical parameters were within normal limits.

Orthopantamogram showed diffuse radiopacity in the left maxillary sinus region [Figure 3]. Computed tomography revealed a huge cystic mass occupying the left maxilla and the left alveolar process with expansion of the left maxillary sinus, scalloping and displacement of the hard palate on the left side, occlusion of the left nasal cavity [Figure 4], and elevation of the floor of the left orbit [Figure 5].

Based on the past history, current clinical presentation, imaging findings, and the earlier histopathological report, a provisional diagnosis of recurrent aneurysmal bone cyst was made and an incisional biopsy was performed under local anesthesia.

On histopathological examination, the excised tissue showed fibrovascular connective tissue with dense collagen fibres

Access this article online
Quick Response Code:
Website: www.jomfp.in
DOI: 10.4103/0973-029X.86693

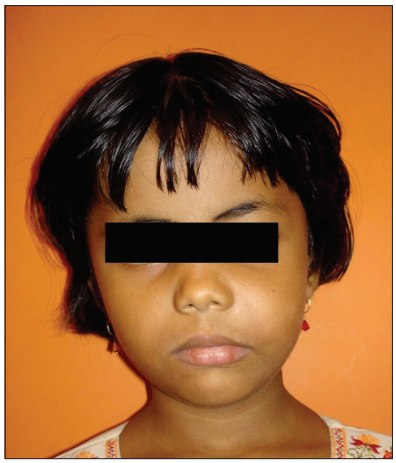

Figure 1: Diffuse swelling over the left middle third of face

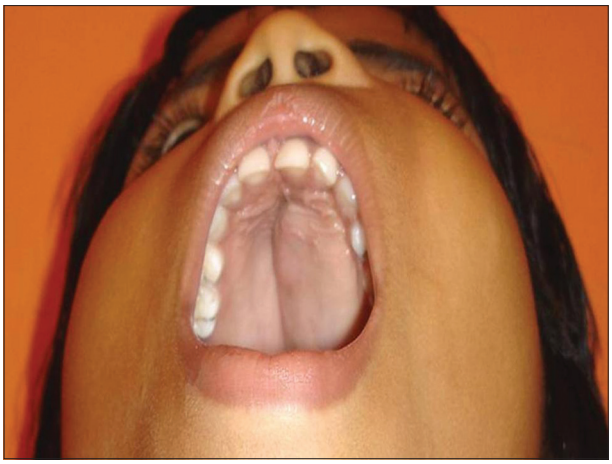

Figure 2: Intraoral swelling seen on the palate

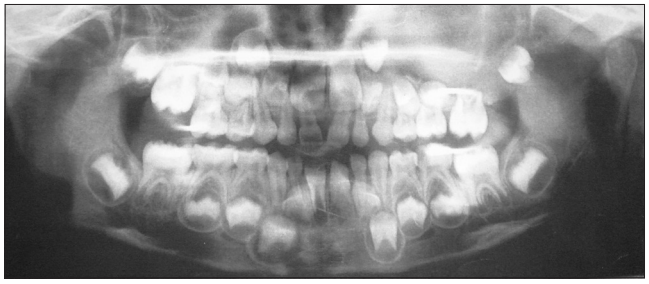

Figure 3: OPG showing diffuse radiopacity in the left maxillary sinus region

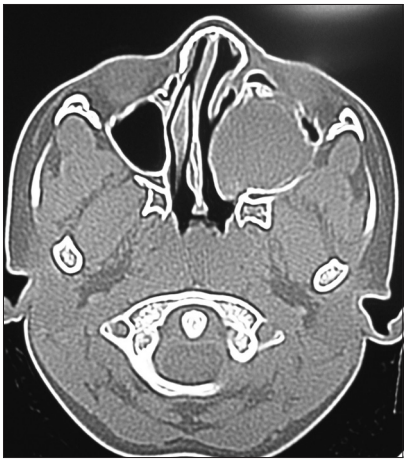

Figure 4: Axial CT showing lesion in the left maxillary sinus with nasal septum deviation

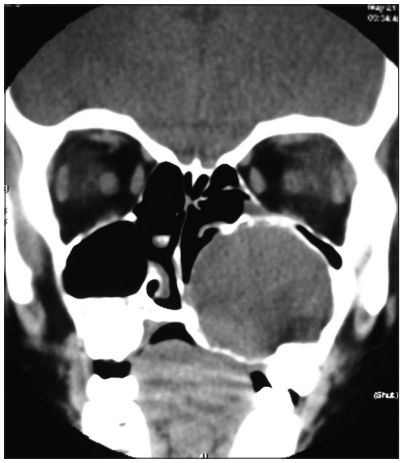

Figure 5: Coronal CT revealing superior displacement of the left orbital floor

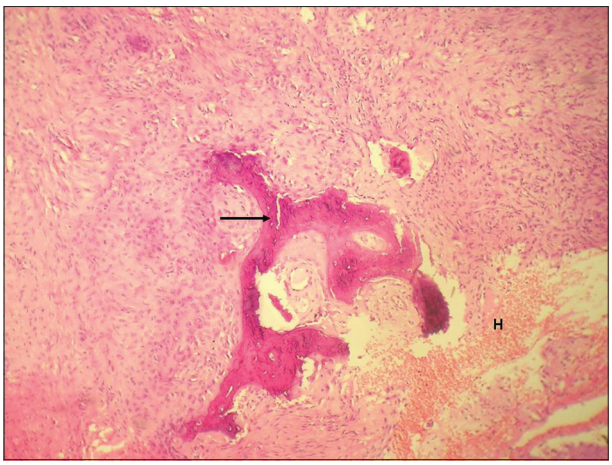

Figure 6: Immature bony trabeculae with peripheral osteoid (arrow) and adjacent area of hemorrhage (H) (H and E, 200×)

interspersed by numerous actively proliferating fibroblasts. Some areas showed parallel arrangement of collagen fibres forming a fibrous capsule. Beneath this layer, areas of immature bony trabeculae with osteoblastic rimming and osteoid formation were seen [Figure 6]. Some areas showed cholesterol clefts with multinucleated giant cells. In the periphery of the lesional tissue, areas of dystrophic calcification were seen. In addition, sections from the central areas of the lesion showed blood filled spaces of varying sizes [Figure 7] and giant cells [Figure 8]. These features were collectively suggestive of trabecular variant of juvenile ossifying fibroma with secondary aneurysmal bone cyst like changes. Due to the rapid growth and recurrent nature of the lesion, surgical resection was performed under general anesthesia. The patient recovered well and has been asymptomatic since 1 year.

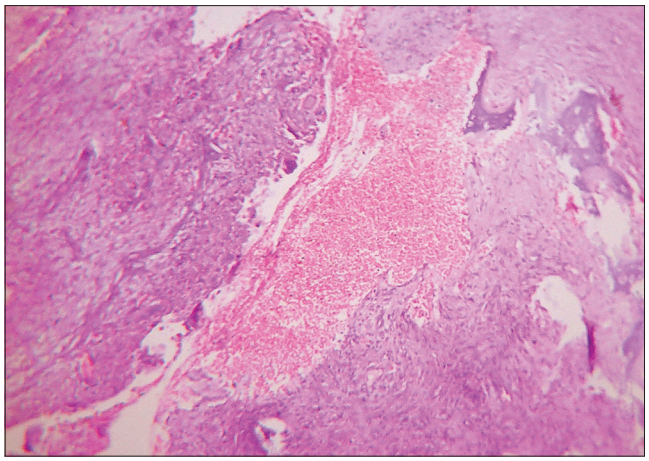

Figure 7: Section showing blood filled cavity (H and E, 200×)

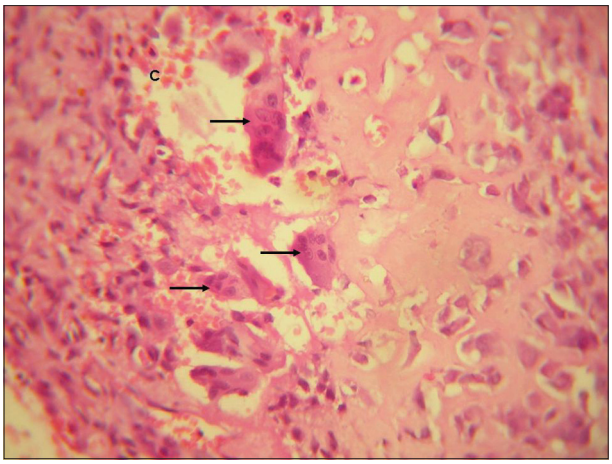

Figure 8: Section showing multinucleated giant cells (arrow) close to extravasated red blood cells (C) (H and E, 400×)

DISCUSSION

The term juvenile (aggressive) ossifying fibroma was used in the second edition of the WHO classification of odontogenic tumors.[13] Previously it was known under different terms such as osteoid fibroma with atypical ossification,[14] juvenile active ossifying fibroma,[12] psammomatous desmo-osteoblastoma and trabecular desmo-osteoblastoma.[7] In 2002, El-Mofty classified the lesion based on histopathologic features as trabecular juvenile ossifying fibroma and psammomatoid juvenile ossifying fibroma.

Our case was diagnosed as trabecular variant of JOF. In a review of case series reported by several authors between 1965 and 2002[2,7-9,11,15,16] the average age of occurrence of TrJOF varied between 2 and 33 years with an average age range of 8.5–12 years.

Maxilla was more commonly involved than mandible. Pain and paresthesia were uncommon. Occasionally, maxillary tumors can lead to nasal obstruction, epistaxis, and eye displacement.

Radiographically, JOF appears radiolucent with variable radio-opaque masses depending on the degree of calcification. The lesion usually has sclerotic rimming which helps in differentiating JOF from fibrous dysplasia. The latter usually has a diffuse border.[17]

Histologically, the TrJOF variant is characterized by cell rich stroma containing spindle-shaped fibroblastic cells with minimal collagen production. Within the stroma there are strands of cellular osteoid along with trabeculae of woven bone. Sometimes, the cellular osteoid is not clearly discernible from the surrounding cell rich stroma[7,9-11,13,16,18]

ABC in JOF develop initially as focal myxoid change in the stroma with hemorrhage and osteoclastic giant cells with gradual expansion and formation of cysts within fibrous walls.[2]

Secondary ABC-like areas in JOF have been reported in earlier literature. Most of such cases are associated with psammomatoid variant of JOF.[4-8] ABC in trabecular variant of JOF is far less common.

Out of 69 cases of PsJOF reviewed by Makek, 3 cases showed ABC like areas. In the same series, out of 24 TrJOF cases, not a single case of secondary ABC was reported.

However cases of TrJOF with areas of cystic degeneration and ABC changes have been reported by other authors.[9-11] Our case had features of TrJOF with areas of secondary ABC.

Nofkke[10] reported a case of a 4-year-old boy who was provisionally diagnosed with JOF. The boy did not undergo any treatment at that time, but returned 7 years later with a considerably enlarged swelling. Postsurgical tissue examination revealed areas of TrJOF with ABC.

Another case of ABC arising in an area where a cementifying fibroma was excised earlier was reported by Robinson in 1985.[19]

Rapid growth is usually associated with younger age group and when associated with secondary ABC.[2,12]

In our case the earlier incisional biopsy and histopathalogical examination had revealed definitive features of aneurysmal bone cyst only. Subsequent histopathological examination of multiple sections from the second biopsy showed features of both aneurysmal bone cyst and juvenile ossifying fibroma.

This finding was also confirmed by examining sections from different areas of the resected specimen.

Through this report we would like to emphasize the need for thorough histopathological examination of post-surgical specimens in cases that show rapid growth and recurrence potential for confirmation of the earlier diagnosis. This report also goes along with theory on the pathogenesis of aneurysmal

bone cyst that the cyst may arise as a secondary change in a pre-existing primary lesion.

REFERENCES

1. Sun G, Chen X, Tang E, Li Z, Li J. Juvenile ossifying fibroma of the maxilla. Int J Oral Maxillofac Surg 2007;36:82-5.
2. El-Mofty S. Psammomatoid and trabecular juvenile ossifying fibroma of the craniofacial skeleton: Two distinct clinicopathologic entities. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002;93:296-304.
3. Struthers PJ, Shear M. Aneurysmal bone cyst of jaws. II. Pathogenesis. Int J Oral Surg 1984;13:92-100.
4. Nasser MJ. Psammomatoid ossifying fibroma with secondary aneurysmal bone cyst of frontal sinus. Childs Nerv Syst 2009;25:1513-6.
5. Blayney AW, el Tayeb AA. The hybrid fibro osseous lesions. J Laryngol Otol 1986;100:291-302.
6. Margo CE, Ragsdale BD, Perman KI, Zimmerman LE, Sweet DE. Psammomatoid (juvenile) ossifying fibroma of the orbit. Ophthalmology 1985;92:150-9.
7. Makek M. Clinical pathology of fibroosteo-cemental lesions of the craniofacial skeleton and jaw bones. Basel (Switzerland): Karger; 1983. p. 128-227.
8. Slootweg PJ, Panders AK, Nikkels PG. Psammomatoid ossifying fibroma of the paranasal sinuses. An extragnathic variant of cemento-ossifying fibroma: Report of 3 cases. J Craniomaxillofac Surg 1993;21:294-97.
9. Slootweg PJ, Muller H. Juvenile ossifying fibroma. Report of four cases. J Craniomaxillofac Surg 1990;18:125-9.
10. Noffke CE. Juvenile ossifying fibroma of the mandible. An 8 year radiological follow-up. Dentomaxillofac Radiol 1998; 27:363-6.
11. Williams HK, Mangham C, Speight M. Juvenile ossifying fibroma. An analysis of 8 cases and a comparison with other fibro osseous lesions. J Oral Pathol Med 2000;29:13-8.
12. Johnson LC, Yousefi M, Vinh TN, Heffner DK, Hyams VJ, Hartman KS. Juvenile active ossifying fibroma: Its nature, dynamic and origin. Acta Otolaryngol Suppl 1991;488:1-40.
13. Kramer IR, Pindborg JJ, Shear M. Histological typing of odontogenic tumors. 2nd ed. Berlin: Springer-Verlag; 1992. p. 28.
14. Benjamins CE. Das osteoid-fibroma mit atypischer Verkalkung im sinus frontalis. Acta Otolaryngol 1938;26:26-43.
15. Dehner LP. Tumours of the maxilla and mandible in children. Clinicopathologic study of 46 histologically benign lesions. Cancer 1973;31:364-84.
16. Reed RJ, Hagy DM. Benign nonodontogenic fibro-osseous lesions of the skull. Report of two cases. Oral Surg Oral Med Oral Pathol 1965;19:214-27.
17. Regezi JA, Sciubba JJ. Oral Pathology: Clinical-Pathologic Correlations. 2nd ed. Philadelphia: PA, Saunders; 1993. p. 205-9.
18. Pindborg JJ, Horting-Hansen E. Juvenile ossifying fibroma. Atlas of diseases of the jaws. Copenhagen: Munksgaard; 1974. p. 68.
19. Robinson PD. Aneurysmal bone cyst: A hybrid lesion? Br J Oral Maxillofac Surg 1985;23:220-6.

How to cite this article: Sankaranarayanan S, Srinivas S, Sivakumar P, Sudhakar R, Elangovan S. "Hybrid" lesion of the maxilla. J Oral Maxillofac Pathol 2011;15:299-302.

Source of Support: Nil. **Conflict of Interest:** None declared.