Juvenile trabecular ossifying fibroma of the mandible

Khurshida Banu¹✉ · Suresh Menon Palikat²

¹ Professor and Head

² Associate Professor

Received: 19 December 2009 / Accepted: 11 February 2010
© Association of Oral and Maxillofacial Surgeons of India 2009

Dept. of Oral and Maxillofacial Surgery,
Vydehi Institute of Dental Sciences and
Research Center, Bangalore

Abstract Juvenile Ossifying Fibroma (JOF) is a rare fibro-osseous neoplasm in young children. This lesion is locally aggressive and spreads quickly. It is usually asymptomatic, achieving a large size. Owing to its aggressive behavior and high recurrence rate, early detection and complete surgical excision are essential. This report details the diagnosis and treatment of a 3-year-old girl presenting with a mandibular swelling that was subsequently determined to be juvenile ossifying fibroma.

Keywords Juvenile ossifying fibroma · Trabecular · Mandible · Neoplasm

Address for correspondence:

Khurshida Banu
Dept. of Oral and Maxillofacial Surgery
Vydehi Institute of Dental Sciences and
Research Center,
No. 82, EPIP Area, Whitefield,
Bangalore, Karnataka, India
E-mail: abdarakib@yahoo.co.in

Introduction

The term juvenile ossifying fibroma is used in naming two microscopically distinct fibro-osseous lesion of the craniofacial skeleton.

Psammomatoid Juvenile Ossifying Fibroma (PSJOF) characterized by small uniform spherical ossicles resembling psammoma bodies. Trabecular Juvenile Ossifying Fibroma (Tr JOF) distinguished by trabeculae of fibrillary osteoid and woven bone.

Psammomatoid Juvenile Ossifying Fibroma (PSJOF) is reported more commonly than Trabecular Juvenile Ossifying Fibroma. It affects patients from wide age range (3 months–72 years) and an older mean age range (16–33 years) as compared with 2–12 years and 8½–12 years respectively for Trabecular Juvenile Ossifying Fibroma [3]. There is a slight male predilection in both types. A significant difference between the two tumours is their site of occurrence. Psammomatoid Juvenile Ossifying Fibroma (PSJOF) occurs predominantly in the sino-nasal and orbital bones, Trabecular Juvenile Ossifying Fibroma predominantly affects the jaws. Aggressive growth occurs in some cases of both the types.

Case report

A 3-year-old female child patient was referred to the Department of Oral and

Fig. 1 Profile view of the lesion

Fig. 2 Postoperative extra-oral front view

Fig. 3 Intraoperative photograph

Maxillofacial Surgery. The patients parents were aware of the right facial swelling over a period of 8 months. The lesion had been

slowly increasing in size since it was noticed. There was no history of trauma, pain, paresis, paresthesia, lymphadenopathy or dysphagia. Physical examination showed a healthy, normal developed young girl with no apparent distress. There was significant facial asymmetry on the right side of the face. Intra-oral examination revealed an approximately 5 x 5 cm mass in the molar, premolar region. The mass was firm and non tender to palpation and not adherent to the overlying skin. There was expansion of buccal cortex and inferior border of mandible. There was no evidence of tooth mobility, abscess formation, dehiscence or malocclusion. Examination of the neck found no abnormalities. A panoramic radiograph revealed a well-circumscribed

radiolucency with faint linear to irregular central opacities. The tooth bud of right mandibular first molar was present. The roots of mandibular right first and second molar were found to be resorbed. Expansion and thinning of the lateral and inferior cortex, and central calcification was evident. A FNAC - Fine needle aspiration of the involved area did not yield fluid. Laboratory values were within normal limits except for a slight increase in Alkaline Phosphatase (ALP).

The lesion was excised under general anesthesia through an intra-oral degloving incision. Inspection of the bony cavity showed no cortical dehiscence. Microscopic examination showed a cellular connective tissue consisting of fibroblasts arranged in a whorled pattern which merged into anastomosing areas of cellular condensation. There were ribbon like osteoid trabeculae lined by plump osteoblasts that showed a transition into areas of woven bone associated with occasional osteoclasts. Clusters of multinucleated giant cells were noted near areas of stromal hemorrhage. Periphery of vital bone was separated by a thick fibrous connective tissue. Based on clinical history and the radiographic and histologic features of the lesion, a diagnosis of Juvenile Ossifying Fibroma Trabecular type was established. The patient was discharged from the hospital and has since been on follow-up, with no evidence of recurrent disease after 8 months.

Discussion

The term Ossifying Fibroma has been used to describe clinically and microscopically heterogeneous lesions of the skull [15]. Juvenile Ossifying Fibroma is used in the literature to describe 2 distinct histopathologic variants of ossifying fibroma of the craniofacial skeleton [1,2]. These are referred to as Psammomatoid Juvenile Ossifying Fibroma (PsJOF) and Trabecular Juvenile Ossifying Fibroma (TrJOF) [3,4]. Although Juvenile Ossifying Fibroma can occur anywhere in the skeleton, its highest incidence is in the facial bones. Most commonly the maxilla [4]. Mandibular and extra cranial involvement are rare [5,6,8,10,11,12,20].

Table 1 summarizes the three clinic pathologic variants of ossifying fibroma with their key features.

Trabecular Juvenile Ossifying Fibroma is characterized by a progressive and

Fig. 4 OPG showing mixed radiopaque and radiolucent lesion

Fig. 5 OPG showing immediate postoperative

Fig. 6 Postoperative OPG after 10 months

sometimes rapid aggressive growth [3,4]. The tumour expands the affected bone leading to facial asymmetry.

Radiographically, it is expansive, well defined unilocular or multilocular with cortical thinning and perforation [5,6,7]. The tumour mass is radiolucent with variable calcification – Induced Radio opacities. Occasionally producing a ‘Ground Glass’ appearance [8]. Root resorption and displacement of involved teeth are observed. In the mandible the angle and ramus are more

commonly involved [5,9]. Trabecular Juvenile Ossifying Fibroma is not encapsulated, but is separated from surrounding bone by a radiopaque border, and this finding can help in differentiation from fibrous dysplasia [7,8,17].

Microscopic examination of Trabecular Juvenile Ossifying Fibroma shows a well defined but unencapsulated lesion that infiltrates surrounding bone [15,16,17]. The tumour is composed of cell-rich fibroblastic spindle cell stroma. Fibrillary osteoid matrix

Table 1 Summarizes the three clinic pathologic variants of ossifying fibroma with their key features

	Ossifying fibroma	Juvenile trabecular ossifying fibroma	Juvenile psammomatoid ossifying fibroma
Age (mean)	10–50 years (35)	2–30 years (10)	3 months–72 years (20)
Female:Male	2:1	1.2:1	1.3:1
Site	Tooth-bearing areas Mandible 70% Maxilla 30%	Mandible 50% Maxilla 44% Sinonasal 6%	Sinonasal 62% Maxilla 20% Mandible (ramus) 10% Cranium 8%
Radiology	Well circumscribed Unilocular Variable radiopacity	Well circumscribed Speckled calcifications	Well circumscribed Expands and fills paranasal sinuses – ‘aggressive pattern’ tends to be radiopaque
Histopathology	Cellular fibrous tissue, storiform pattern, separate trabeculae of woven bone, occasional spherical, psammomatoid calcifications, encapsulated	Densely cellular, immature osteoid and in trabecular pattern, osteoblast rimming and cellular osteoid	Densely cellular, spherical cementum-like psammomatoid calcifications, may permeate cancellous bone, myoid areas, giant cells, haemorrhage
Synonyms	Cemento-ossifying fibroma Cementifying fibroma Periodontoma	Juvenile aggressive ossifying fibroma Juvenile ossifying fibroma WHO type Trabecular desosteoblastoma	Cemento-ossifying fibroma Juvenile ossifying fibroma with psammomatoid ossicles Juvenile ossifying fibroma psammous desosteoblastoma

H&E picture of the lesion at 10x view arrows indicating the immature trabeculae of bone

develops incorporating plump eosinophilic osteoblastic cells. Progressive calcification of the osteoid results in anastomosing trabeculae of immature woven bone and focal collections of multinucleated giant cells are not seen. Not all cases show a fast and aggressive growth rate [5,6]. Surgical excision is the recommended treatment for Trabecular Juvenile Ossifying Fibroma

H&E picture of the lesion at 40x view with black arrow indicating the immature trabeculae of bone and light black arrow indicating the cellular connective tissue

[6,9]. Because of infiltrating borders, the tumour cannot be shelled out but should be conservatively excised entirely. Recurrences are seen in 30% to 58% [2,7,10] of cases. More than one excision may be required to achieve a cure [7]. No malignant transformation has been reported and radical surgical procedures are not necessary [3,6] which corroborates with the treatment

provided in the present case. The age of the child and the growth considerations also mitigated against a radical surgical approach.

Conclusion

The incidence of JOF is still unknown because of the relatively few cases reported till date. In the case discussed here, clinical, radiological and histological features confirmed the diagnosis of JOF.

The clinical management of JOF remains uncertain. Small lesions can be treated conservatively by curettage or enucleation [3,7] and follow-up for a prolonged period of time is recommended [5] as there is high rate of recurrence of these tumours as described in the literature [3,4,7].

References

1. Waldron CA (1993) Fibro-osseous lesions of the jaws. J Oral Maxillofac Surg 51(8): 828–835

2. Slootweg PJ, Panders AK, Koopmans R, Nikkels PG (1994) Juvenile ossifying fibroma. An analysis of 33 cases with emphasis on histopathological aspects. *J Oral Pathol Med* 23(9): 385–388
3. El-Mofty S (2002) Psammomatoid and Trabecular Juvenile ossifying fibroma of the craniofacial skeleton. Two distinct clinico pathologic entities. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 93(3): 296–304
4. Johnson LC, Yousefi M, Vinh TN, Heffner DK, Hyams VJ, Hartman KS (1991) Juvenile active ossifying fibroma. Its nature, dynamics and origin. *Acta Otolaryngol Suppl* 488: 1–40
5. Noffke CEE (1998) Juvenile ossifying fibroma of the mandible. An 8 year radiological follow-up. *Dentomaxillofac Radiol* 27(6): 363–366
6. Rinaggio J, Land M, Cleveland DB (2003) Juvenile Ossifying Fibroma of the mandible. *J Pediatric Surg* 38(4): 648–650
7. Shekhar MG, Bokhari K (2009) Juvenile Aggressive Ossifying Fibroma of the Maxilla. *J Indian Soc Pedod Prev Dent* 27(3): 172–174
8. Thankappan S, Nair S, Thomas V, Sharafudeen KP (2009) Psammomatoid and Trabecular variants of Juvenile Ossifying Fibroma – 2 case reports. *Indian J Radiol Imaging* 19(2): 116–119
9. Gardner JA, Hanft RJ (1960) An Ossifying Fibroma of the mandible. Report of a case. *Oral Surg Oral Med Oral Pathol* 13: 1288–1294
10. Slootweg PJ, Moller H (1990) Juvenile Ossifying Fibroma. Report of 4 cases. *J Craniomaxillofac Surg* 18(3): 125–129
11. Halkias LE, Larsen PE, Allen CM, Steinberg MJ (1998) Rapidly growing, Expansive mass of the Mandible in a 6 year old boy. *J Oral Maxillofac Surg* 56(7): 866–871
12. Sciubba JJ, Younai F (1989) Ossifying Fibroma of the mandible and maxilla. Review of 18 cases. *J Oral Pathol Med* 18(6): 315–321
13. Makek MS (1987) So called ‘Fibro Osseous Lesions’ of Tumourous origin. Biology confronts terminology. *J Craniomaxillofac Surg* 15(3): 154–167
14. Slootweg PJ (1996) Maxillofacial Fibro-Osseous lesions, Classification and differential diagnosis. *Semin Diagn Pathol* 13(2): 104–112
15. Pefer A, Reichart / Hans P. philipsen Odontogenic Tumours and allied lesion. 2004 - Q.P. London. EDS 278–279
16. Waldron CA Bone Pathology. In : Neville BW, Damm DD, Allen CM, Bouquot JE, Editors (1995) *Oral and maxillofacial Pathology* Philadelphia: WB Saunders 443–492
17. Williams HK, Mangham C, Speight PM (2000) Juvenile Ossifying Fibroma; An analysis of Eight cases and a comparison with other Fibro-Osseous lesions. *J Oral Pathol Med* 29(1): 13–18
18. Zama M, Gallo S, Santecchia L, Bertozzi E, DE Stefano C (2004) Juvenile Active Ossifying Fibroma with massive involvement of the mandible. *Plast reconstr Surg* 113(3): 970–974
19. Saiz-Pardo-Pinos AJ, Olmedo-Gaya MV, Prados-Sanchez E, Vallecillo-Capilla M (2004) Juvenile Ossifying Fibroma: A case study. *Med Oral Pathol Oral Cir Bucal* 9(5): 456–458
20. Paul M Speight, Roman Carlos, *Current Diagnostic Pathology* 2006, issue 12, page 1-10, Mini Symposium, Head & Neck Pathology, Maxillofacial, Fibro Osseous Lesions

Source of Support: Nil, **Conflict of interest:** None declared.