The Saudi Journal for Dental Research (2014) 5, 135–138

King Saud University

The Saudi Journal for Dental Research

www.ksu.edu.sa
www.sciencedirect.com

CASE REPORT

Juvenile psammomatoid ossifying fibroma with secondary aneurysmal bone cyst of mandible☆

S.A. Deshingkar *, S.R. Barpande, J.D. Bhavthankar

Department of Oral Pathology and Microbiology, Government Dental College and Hospital, Aurangabad 431 001, Maharashtra, India

Received 3 October 2012; revised 22 December 2013; accepted 22 December 2013
Available online 16 February 2014

KEYWORDS

Ossifying fibroma;
Juvenile ossifying fibroma;
Psammomatoid juvenile ossifying fibroma;
Aneurysmal bone cyst

Abstract Juvenile psammomatoid ossifying fibroma (JPOF) is a rare, slowly progressive tumor of the extragnathic craniofacial bones, with a tendency towards locally aggressive behavior and recurrence. The pathognomonic histopathologic feature is the presence of spherical ossicles, which are similar to psammoma bodies. Very few cases in association with the secondary aneurysmal bone cyst (ABC) formation have been reported in the literature. Treatment consists of complete surgical removal; the incomplete excision has been associated with a high local recurrence rate. The prognosis is good because malignant change and metastasis have not been reported. We report a case of JPOF of the mandible with secondary ABC in an 18-year-old male patient.

1. Introduction

Juvenile ossifying fibroma (JOF) is a fibro-osseous lesion that occurs in the facial bones.[1] It is also called aggressive ossifying fibroma due to its aggressiveness and high tendency to recur. Due to its distinct histological features, it is recognized as a separate histopathological entity among the fibro-osseous group of lesions.[1–3] El-Mofty identified two histopathological variants; juvenile trabecular ossifying fibroma (JTOF) and juvenile psammomatoid ossifying fibroma (JPOF).[4]

JPOF is a rare tumor of the extragnathic craniofacial bones. It is a slowly progressive lesion with a tendency to invade surrounding tissue, and recur after surgical excision. The pathognomonic histopathologic feature is the presence of spherical ossicles, which are similar to psammoma bodies.[4] Here we report a case of JPOF of the mandible associated with aneurysmal bone cyst (ABC) in an 18-year-old male patient. To our knowledge, very few cases of JPOF of the mandible in association with ABC have been reported in the literature.[5–11]

2. Case report

An 18-year-old male presented with a painless swelling on the right side of his face since 8 months. Extra-orally the

* Corresponding author. Tel./fax: +91 0240 2402383; mobile: +91 97629 80179.
E-mail addresses: sanket_dr2005@rediffmail.com (S.A. Deshingkar), sureshbarpande@rediffmail.com (S.R. Barpande), jdbhavthankar@gmail.com (J.D. Bhavthankar).

☆ This case report has been presented as a scientific poster at "Post Graduate Convention of Indian Association of Oral and Maxillofacial Pathologist", held at Pune, India during July 2012.

Peer review under responsibility of King Saud University.

Production and hosting by Elsevier

http://dx.doi.org/10.1016/j.sjdr.2013.12.002

swelling extended from para-symphyseal region to the posterior border of the ramus of mandible and superioinferiorly from the preauricular region to the inferior border of the body of mandible (Fig. 1A). Overlying skin was normal. No cervical lymphadenopathy was noted and mouth opening was normal. Intra-oral examination revealed a swelling in retromolar region with obliteration of lower buccal vestibule from distal of left mandibular first premolar to the anterior border of ramus of the mandible (Fig. 1B). The labial cortex was expanded with no apparent mucosal changes. On palpation, the swelling was hard in consistency with no fluctuation elicited.

An orthopantomogram showed a multilocular expansile lesion on the left side of body and ramus of the mandible without invasion of the coronoid and condylar processes (Fig. 1C). The lesion had in part wispy radiopacities, with endosteal scalloping and a narrow transitional zone with the adjacent normal bone. Routine hematological and urine investigations ascertained values within normal limits.

Histopathological examination of incisional biopsy revealed a cellular proliferation of uniform, cytomorphologically bland fibroblastic spindle cells with no evidence of mitotic figures (Fig. 2A). Interspersed between the fibroblastic cells were numerous irregular and spherical ossicles exhibiting varying degrees of calcification with minimal extra-cellular collagen deposition. These psammoma body-like ossicles were relatively acellular, with peripheral eosinphillic rimming and distributed homogeneously throughout the section (Fig. 2B). Large vascular and sinusoidal spaces devoid of the epithelial lining and engorged with red blood cells (Fig. 3A) were evident. Osteoclastic giant cells were focally demonstrated in the lining of these spaces establishing the presence of associated ABC (Fig. 3B). The constellation of histomorphologic, radiographic and clinical features of this lesion supported an interpretation of JPOF with secondary ABC.

Due to patient's poor economic status, a primary reconstruction with a bone graft could not be done. Standard hemi-mandibulectomy with disarticulation of the condyle was done. A titanium reconstruction plate with condylar head which was pre-adapted on cadavaric mandible was prepared to give the best patient profile. Arch bars were fixed to the maxilla and right mandible and maxillo-mandibular fixation (MMF) was done. The reconstruction plate was fixed with three screws in the anterior mandible. The MMF was released and condylar movement was observed. Suture removal was done after 8 days and MMF was done with elastics for 3 weeks to maintain the occlusion.

The post-operative facial appearance and oral function was satisfactory. Due to the economic reason patient refused further reconstructive surgery and did not turn up for follow-up.

3. Discussion

Slootweg et al.[12] identified two distinct groups, the JOF-WHO type and JOF-Psammomatoid type, based on the age of occurrence: the mean age of for JOF-WHO is 11.8 years and for JOF Psammomatoid type is 22.6 years.[6] El-Mofty suggested two categories, JTOF and JPOF, based on histopathologic criteria.[4] The two categories have a distinct predilection for specific age-groups: the average age for JTOF is 81/2–12 years, whereas that for JPOF is 16–33 years.[4,13] Recently, non-random chromosome break points at Xq26 and 2q33 resulting in (X;2) translocation have been identified.[13] Unfortunately, no studies have been performed on JTOF which would enable the identification of cytogenetic differences between two variants.

The age of the patients ranges from 3 months to 72 years, with mean age of occurance 17.7 years.[6,7,13] It is most commonly seen in first and second decade of life.[4,6–8] It shows slight male predominance with the male:female ratio of 1.2:1.[4,6–8] The age and gender of the present case is in confinement of the data from literature. Most cases affect sinonasal area and jaws (90%), out of which 10% cases involve mandible.[6,7] Clinical manifestations include proptosis, visual

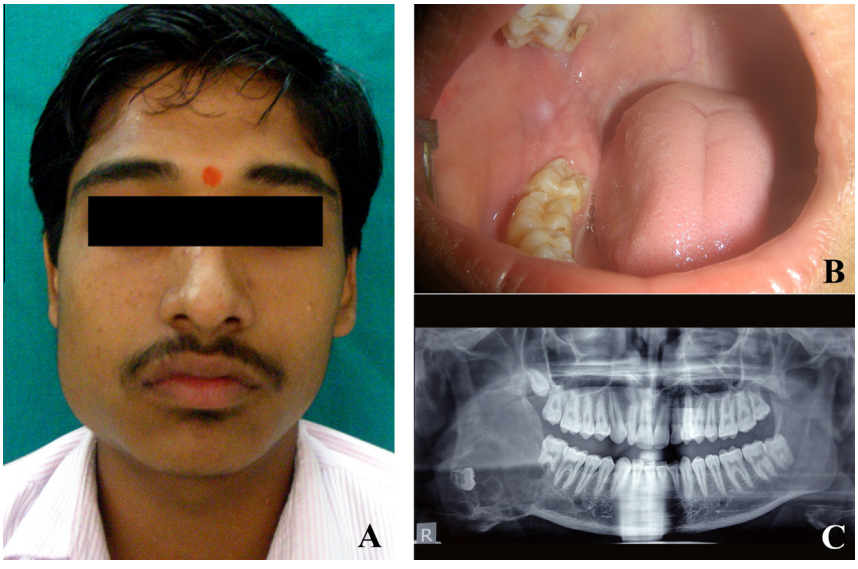

Figure 1 (A) Clinical photographs showing diffuse swelling on left side of the face. (B) Swelling in the left molar region with obliteration of the vestibule. (C) Orthopantomogram showing a multilocular expansile radiolucency involving body, ramus of the left mandible without involvement of condyle and coronoid processes.