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PSAMMOMATOID JUVENILE OSSIFYING FIBROMA IN MANDIBLE: A RARE ENTITY. REVIEW OF LITERATURE AND REPORT OF A CASE

Surgery

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ABSTRACT

Psammomatoid juvenile ossifying fibroma (PsJOF) is a rare benign fibro-osseous tumor with slowly progressive and aggressive local growth leading to invasion and destruction of the surrounding tissue, bone erosion and high chances of recurrence after surgical excision. Its age of onset, clinical picture and Zealous behavior makes it different from the other fibro-osseous lesions of the jaw. Histopathological diagnosis of PsJOF is an enigma even for a pathologist. 70% of PsJOF is reported in the paranasal sinuses, 20% in the maxilla, and only 10% in the mandible. Here we report an additional case of PsJOF causing an asymptomatic expansile swelling of right mandibular body region in a 15 year old young male patient.
Histopathologically, large number of spheroidal cementoid ossicles merging into the surrounding stroma accompanied by spheroidal to trabecular osteoid tissue were seen. Unlike resection which is widely reported in literature as the preferred treatment modality, we chose to do surgical excision and peripheral osteotomy and 2 year follow up shows no signs of recurrence.
Our Purpose is to review and compare the demographic, clinical, and histopathologic features of published cases of PsJOF.
In summary, surgeons should develop the surgical plan according to the age of occurrence, growth status, extent of the lesion and relapse status.

KEYWORDS

Juvenile ossifying fibroma; Psammomatoid; Fibro osseous lesion; Aggressive.

Introduction

Fibro-osseous lesions of the craniofacial skeleton represent a group of benign conditions that are characterized by replacement of normal bone with fibrous connective tissue that gradually undergoes mineralization [1]. Among them are ossifying fibroma, fibrous dysplasia, and osseous dysplasia. In which, True neoplastic features are represented by ossifying fibroma (OF) [2].

OF can be further divided into conventional and juvenile forms (JOF) [3]. JOF is further classified into two distinct entity as Psammomatoid juvenile ossifying fibroma (PsJOF) and trabecular juvenile ossifying fibroma (TrJOF) by El- Mofty in 2002 [4].

The average age of incidence of PsJOF according to different studies ranges from 16 to 33 years with age range of 3 months to 72 years [5]. PsJOF occurs predominantly in the sinonasal complex and orbital bones. Among all bones of maxillofacial regions, mandible is rarely affected [6].

PsJOF is clinically characterized as a progressive disorder and sometimes shows rapid expansion of the affected area. Patients may develop proptosis, exophthalmos, and bulbar displacement if the orbital bones and paranasal sinuses are involved. In mandibular involvement; Paresthesia, Cortical disruption, tooth displacement and very rarely root resorption can be seen [7]. A ground glass appearance with well-defined osteolytic lesionis seen radiographically [8]. The lesion may range in size from 2 to 8 cm in diameter and may appear multiloculated on Orthopantomogram or computedtomography (CT) scans [9]. Histopathology shows dense cellular fibrous stroma and characteristic spheroidal calcifications called **Psammoma bodies** [10].

The management of PsJOF remains controversial. It has a very strong propensity to recur (Recurrence rate: 30-58%) [11] as compared to trabecular. Hence, it becomes very important as well as difficult to decide a more acceptable treatment plan which can help for better prognosis and quality of life. The treatment can be carried out both ways, conservative or radical. But the treatment plan should be decided by keeping in mind about the nature and involvement of the lesion with surrounding area [12].

Some surgeons prefer local radical surgery owing to the aggressive behavior and high propensity for its recurrence [4,12] whereas others consider conservative treatment. The reason being beneficial for the young patients in account of their growth and development, the esthetic appearance and preservation of nerve function [13].

In our study we are reporting an unusual case of PsJOF which is present in the body of right mandible, along with a comprehensive literature review regarding clinical, histologic, and radiographic features with its prognosis, and treatment for PsJOF of the jaws.

Case report

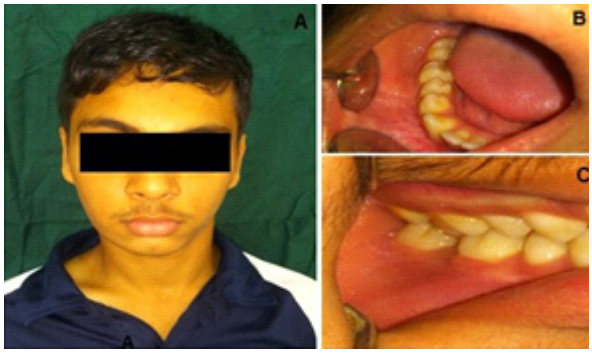
A 15-year-old boy presented with a painless progressive swelling on the right side of his face since 4 month.

Extra-orally, swelling was present just behind the corner of the mouth to 2 cm anterior to posterior border of ramus of mandible (Fig. 1A).

Intra-orally, swelling was present distal of the right mandibular first premolar to the right mandibular second molar. Lower buccal vestibule

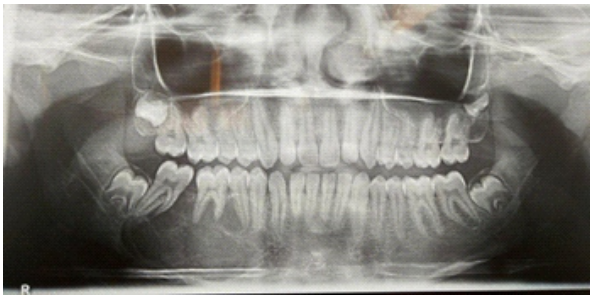
was obliterated (Fig. 1B, C). On palpation, the buccal and lingual cortices were expanded and the swelling was painless, hard in consistency with no fluctuation elicited.

Fig. 1. Clinical photographs showing (A) diffuse swelling on right side of the face; (B,C) swelling in the right molar region with obliteration of the vestibule.



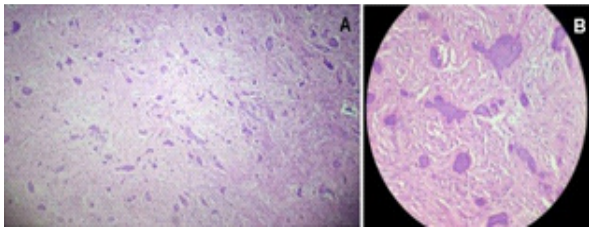
An Orthopantomogram showed a multilocular expansile lesion on the right side of the body of the mandible (Fig. 2) with endosteal scalloping and a narrow transitional zone with adjacent normal bone. Inferior border of the body of mandible was intact.

Fig. 2. Orthopantomogram shows a well-defined multilocular radiolucency involving body of the right mandible



Histopathological examination (H & E stain) of incisional biopsy revealed a cell-rich connective tissue containing numerous acellular spheroidal cementoid ossicles with basophilic concentric laminations and osteoid rim, characterizing Psammoma bodies. Trabeculae of woven bone and irregular osteoid were also observed (Fig. 3A). The fibroblastic stroma consisted of spindle-shaped cells arranged in strands and whorls (Fig. 3B). Blood vessels and small hemorrhagic areas were also noted. So, clinical, histological and radiographical features of this lesion supported an interpretation of PsJOF.

Fig. 3. Histopathological examination showing (A) numerous acellular spheroidal cementoid ossicles with basophilic concentric laminations and osteoid rim, characterizing Psammoma bodies. (B) Fibroblastic stroma consisted of spindle-shaped cells arranged in strands and whorls; (H and E stain)



Tumor was removed in multiple pieces and involved teeth were also extracted (Fig. 4). Inferior alveolar nerve was preserved and vigorous curettage followed by peripheral osteotomy was done. At 2 year postoperatively, a panoramic radiograph showed considerable neoformation of bone (Fig. 5B).

Fig. 4. Gross appearance of tumor. It showed multiple pieces of tumor which was yellowish white in color and gritty in consistency.

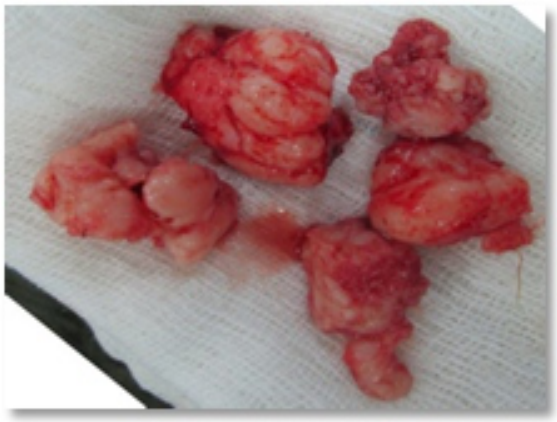
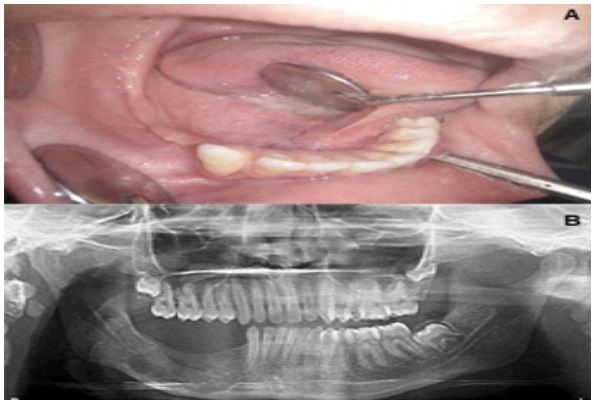


Fig. 5. Follow up after 2 year Post-operatively (A) Intra oral picture showing uneventful normal healing and (B) Orthopantomogram showed neoformation of bone.



Review of literature

74 cases of PsJOF are documented in the English literature review from the year 1985 till 2017. PubMed database was used to conduct the literature review using the term 'Psammomatoid juvenile ossifying fibroma'. 5 reviews and 37 case reports regarding this topic was the statistics obtained. Out of 74 cases, only 32 cases were restricted to the jaws: 11 in the maxilla and 21 in the mandible.

Table -1 summarizes the data of the cases affecting the jaws which includes the present case.14 males and 17 females had manifestations of the disease presenting in jaws giving a male-to-female ratio of 1.2:1. The average age was 17 years with a range of 4 years to 46 years with mean female age of 20.8 years and mean male age of 13 years. Mandible to maxilla ratio was 1.9:1. Predilection for the right side of jaws involvement was seen in 17(58%) cases. Site predilection in the mandible was for body i.e.86% of total cases whereas 14% affected the ramus (Table 1).

Table 1: Analysis of literature review

Jaw affected	Number of Cases	Male	Female	Right side	Left side	Body involvement (%)	Ramus involvement (%)
Maxilla	11	3	8	6	3	-	-
Mandible	21	12	9	11	10	86	14
Mean age (in year)	-	13	20.8	-	-	-	-

Radiographic examination of 10(38%) cases showed radiolucent lesions, 18(62%) case showed a mixed radiodensity. 13(45%) were multilocular radiolucent lesions and 16(55%) cases showed unilocular radiolucent area. Cortical expansion was reported in 79% of cases. Various treatment modalities were used to treat the patients. In which, 14(56%) cases were treated by resection and reconstruction, 4(16%) cases by total or partial maxillectomy with reconstruction and 7(28%) cases by enucleation or excision with curettage. 16 cases were compliant for follow up visits. The available data is of an average of 29 months follow up with range of 2 months to 128 months. Recurrence was not seen in any of the cases (Table 2).

Table 2: Analysis of the Characteristic features of Cases of PsJOF of the Jaws

Case No.	Year	Authors	Location	Gender	Age year	Radiographic features	Treatment	Follow up. (month)	Recur
1	2002	El. Mofty et al4	Left Mandibular Ramus	Male	15	Mixed/Multilocular/Cortical Expansion	Segmental Resection with Reconstruction	12	No
2	2007	Foss et al8	Right Mandibular Body and Ramus	Male	4	Mixed/Unilocular/Cortical Expansion	Hemimandibulectomy	-	-
3	2009	Smith et al16	Right Mandibular Body	Female	12	Mixed/Multilocular/Cortical Expansion	Segmental Resection with Reconstruction	16	No
4	2009	Thankappan et al22	Left Mandibular Body	Female	27	Radiolucent/Multilocular /Cortical Expansion	Segmental Resection with Reconstruction	-	-
5	2011	Patigaroo et al23	Right Maxilla	Female	20	Radiolucent/Unilocular/ Cortical Expansion	Total Maxillectomy with STSG	-	-
6	2011	Malathi et al24	Case 1: Right Maxilla Case 2: Left Mandibular Body	Female Female	46 31	Mixed/Unilocular/Cortical Expansion Mixed/Unilocular/Cortical Expansion	Partial Maxillectomy -	- -	- -
7	2011	Maria et al25	Left Mandibular Body	Female	17	Radiolucent/Unilocular/ Cortical Expansion	Enucleation with Curretage	6	No
8	2012	Tolentino et al26	Case 1: Right Mandibular Body Case 2: Right Mandibular Body	Male Male	12 20	Radiolucent/Multilocular /Cortical Expansion Radiolucent/Multilocular /Cortical Expansion	Excision and Curretage with Reconstruction -	15 -	No -
9	2012	Rao et al27	Right Mandibular Body and Right and Left Anterior Mandible	Female	14	Mixed/Multilocular/Cortical Expansion	Segmental Resection with Reconstruction	12	No
10	2013	Halama et al28	Left Maxilla	Female	7	Mixed Radiolucent-Radioopaque/Unilocular/ Cortical Expansion	Left Maxillectomy with Reconstruction	-	-
11	2013	Ranganath et al29	Left Maxilla and Maxillary Sinus	Male	14	Radiolucent/Unilocular	Segmental Resection with Reconstruction	-	No
12	2013	Guttikonda et al30	Left Maxilla and Maxillary Sinus	Female	20	Mixed/Unilocular/ Cortical Expansion	Left Maxillectomy	-	No
13	2013	Patil et al31	Right Mandibular Ramus	Male	7	Mixed/Unilocular/Cortical Expansion	Surgical Excision with Curretage	36	No
14	2013	Yadav et al32	Right Mandibular Body and Right and Left Anterior Mandible	Female	14	Mixed/Multilocular/Cortical Expansion	Segmental Resection with Reconstruction	6	No
15	2013	Leonardo et al1	Left Mandibular Body and Angle	Male	4	Radiolucent/Multilocular /Cortical Expansion	Osteotomy with Cryotherapy	18	No
16	2013	Melo et al33	Entire Mandible	Female	35	Mixed/Multilocular/Cortical Expansion	Mandibulectomy with Reconstruction	-	-
17	2014	Tamgudge et al34	Left Mandibular Body and Ramus	Male	7	Mixed/Multilocular/Cortical Expansion	Enucleation with Curretage	2	No
18	2015	Gantalu et al35	Right Maxilla	Male	15	Mixed/Unilocular/Cortical Expansion	Enucleation	-	No
19	2015	Shruthi et al36	Left Mandibular Body and Ramus	Male	30	Radiolucent/Multilocular /Cortical Expansion	Segmental Resection with Reconstruction	6	No
20	2015	Kumar et al37	Right Mandibular Body	Male	20	Mixed/Unilocular/Cortical Expansion	Segmental Resection with Reconstruction	-	-
21	2015	Han et al12	Case 1: Left Mandibular Body Case 2: Right Mandibular Body Case 3: : Right Mandibular Body Case 4: Right Maxilla Case 5: Right Maxilla	Female Male Female Female Female	19 13 16 19 15	Mixed/Unilocular Mixed/Unilocular Mixed/Unilocular Radiolucent/Multilocular Mixed/Unilocular	Segmental Resection with Reconstruction Curretage Segmental Resection with Reconstruction Segmental Resection with Reconstruction Segmental Resection	87 20 95 128 13	No Yes No No No
22	2016	Manjunatha et al 38	Right Mandibular Body and Ramus	Male	6	Radiolucent/Multilocular /Cortical Expansion	Enucleation	3	No
23	2017	Present case	Right Mandibular Body	Male	15	Radiolucent/Multilocular /Cortical Expansion	Surgical Excision with Curretage and Peripheral Osteotomy	24	No

Discussion

The ambit of fibro-osseous lesions is difficult to understand because it is too wide. It includes a number of different developmental, dysplastic or reactive and neoplastic lesions like fibrous dysplasia, OF, JOF, and others [14]. Among all the variants of JOF, PsJOF is a unique variant which has a predilection for the sinonasal tract and the orbit is always present at the center on the periorbital, frontal, and ethmoid bones [6].
It is necessary for a clinician to understand the disease in order to perform a diagnosis, proper treatment plan and to predict the prognosis accurately. There is enormous resemblance between JOF and benign fibro-osseous lesions. They possess similarity in relation to clinical features, radiographic features and histopathological also. Hence, it is of immense importance for a clinician to thoroughly understand the disease.

It develops primarily within the craniofacial bones of children, which is the reason behind the lesion to be termed as “juvenile”, although there are some cases of JOF which have been also reported in adult patients [15].

These mesodermal jaw tumors commonly arise from odontogenic cells like periodontal ligament or from primitive mesenchymal cell nest [14].

Maxilla was described as the region where tumors occur the most by Slootweg et al [10]. For Brannon and Fowler [16], the mandible was the predominant site. However, Johnson et al [17] affirm that 90 % of the lesions occur in the paranasal sinuses and 10 % in the mandible. In our literature review, we observed the mandible: maxilla ratio is 1.9:1 thus showing the mandible being the prominent site.

According to 2005 WHO definition, the age at onset of JOF is 15 years. However, According to El-Mofty, the average age of occurrence of the trabecular type is 8.5 to 12.0 years, whereas that of PsJOF is 16.0 to 33.0 years [4]. The onset of JOF can be seen as early as 3 months and can range up to 72 years as reported by Johnson et al [17]. In general, patients with PsJOF are a few years older than those with TrJOF.

Gender predilection has always been a matter of controversy with some authors claiming predilection for either sex whereas Johnson et al. found higher incidence [17] in females and El-Mofty [4] reported a male predilection. In our analysis, male: female ratio is 1.2:1 including present case.

Radiographically, the lesions can be radiolucent, mixed or radiopaque, depending on the degree of mineralization. According to our search, most of the PsJOF cases affecting the jaws presented as radiolucent areas with irregular radiopaque areas, as reported in the present case.

In present case, we considered odontogenic Keratocystic tumor, odontogenic myxoma, ossifying fibroma, Ameloblastoma and central giant cell lesion, fibrous dysplasia as deferential diagnosis depending on the clinical and imaging examinations. Moreover, mandibular involvement in the present case and their rarity of occurrence caused a diagnostic dilemma for us.

Fibrous dysplasia evinces a diffuse border whereas PsJOF presents with a well-defined border. Furthermore, fibrous dysplasia retains the overall configuration of the involved bone as compared to PsJOF which has a roughly spherical configuration. There are completely different treatment modalities for both the entity so accurate diagnosis of PsJOF is crucial [15].

The final diagnosis was confirmed after histopathological analysis which showed a cell-rich connective tissue containing numerous acellular spheroidal cementoid ossicles with basophilic concentric laminations and osteoid rim, characterizing **Psammoma bodies**. Fibroblastic stroma consisted of spindle-shaped cells arranged in strands and whorls. Psammoma-Like bodies vary in appearance, but usually have a central basophilic area and a peripheral eosinophilic fringe [18]. These microscopic features were demonstrated in case reported in our study.

The lesion's consistency was predominantly soft with a variable quantity of internal calcifications. On gross examination, the tumor is described as yellowish, white and gritty [19] as seen in our case (Fig.4). There is no standardized treatment plan for PsJOF. More radical treatment was preferred by Toro et al., El-Mofty et al., Smith et al [20,4,16], consisting en bloc resection with rigid internal fixation by reconstruction plates, bone grafts, integrated bone implants, and prosthetic rehabilitation considering the recurrence rate that is 30 to 58%.

To minimize facial deformity and dysfunction and to preserve normal growth, normal esthetic appearance and neural functions in young patients, Conservative treatment modalities should be considered including curettage with peripheral osteotomy, enucleation, and partial resection [13, 21]. Complete resection should be advocate for the lesions which shows aggressive growth, cortical perforation, and the displacement of teeth or root resorption [21].

The surgeons should prefer the conservative treatment in young patient in order to rehabilitate patient's appearance and also to reduce the unfavorable changes in their growth and development.

In our case we opted for a more conservative approach, considering the young age of the patient, his active phase of craniofacial skeleton growth, site of the lesion which enabled easier surgical access, involvement of the surrounding important vital structures, small size of the lesion and intact inferior border of mandible.

In our case, Patient underwent complete surgical excision of tumor mass with peripheral osteotomy and kept on regular follow up at every 2 month.

We considered that our applied therapy was enough to resolve the case, as there is no sign of recurrence after 2 year of follow-up (Fig. 5A, B).

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Conflict of interest: The authors declare that they have no conflict of interest

Ethical approval: All procedures performed in study involving human participant were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent: Informed consent was obtained from human participant included in the study.

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