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Case Report

Dentistry Section

Uncommon Variant of Ossifying Fibroma (Psammomatoid Type) in an Adult Patient: A Case Report

RATHINASAMY KADHIRESAN[1], KUMAR NALIN[2], EKAMBARAM DEEPIKA[3], CYNTHIA JENIFER[4], PRAVEEN AROKIA ANTONY[5]

ABSTRACT

Ossifying Fibroma (OF) is a fibrous neoplasm lesion characterised by various amounts of calcified tissue, such as bone or cementum within a cellular fibrous stroma. Juvenile Ossifying Fibroma (JOF) is a rare benign fibro-osseous neoplasm seen in children and also young adults. The most frequent sites involved sinonasal, orbital, and jaw bones. Based on clinicopathological features, JOF is classified into two distinct variants: Trabecular JOF (TrJOF) and Psammomatoid JOF (PsJOF). The trabecular variant is more commonly seen in the jaw bones and is characterised histologically by trabeculae of immature woven bone within a cellular stroma. In contrast, the psammomatoid variant also referred to as Psammomatoid OF (POF), typically affects the paranasal sinuses, orbit, and craniofacial bones. It is characterised histopathologically by the presence of numerous small, round, psammoma-like ossicles embedded in a densely cellular fibrous stroma. POF is a rare, benign, fibro-osseous variant of OF exhibiting short-term rapid growth and has a high recurrence rate. The diagnosis is based on radiographic and histopathological findings, hereby, the authors present a case report of a psammomatoid variant of OF in right maxillary region in a 26-year-old male patient presented with a mild swelling and pain in the right upper tooth region for past one week. After clinical, radiological, and histological examinations, the diagnosis was confirmed as POF. Surgical excision was performed. There were no signs of recurrence or complications.

Keywords: Central giant cell granuloma, Craniofacial, Fibro-osseous, Psammoma bodies, Sinonasal, Orbital

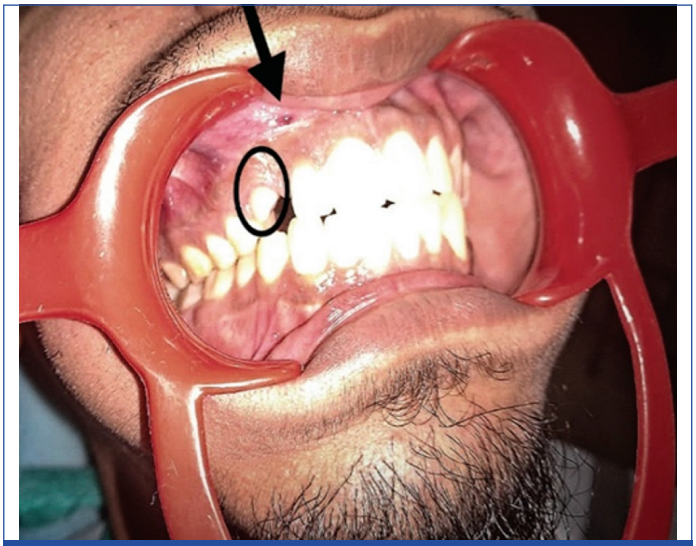
CASE REPORT

A 26-year-old male patient presented to a private dental clinic for his first dental visit with a chief complaint of pain accompanied by mild swelling in the right upper front tooth region, which he had been experiencing for the past one week. The patient denied having any relevant medical history and stated that he was not suffering from any systemic illness, was not on any long-term medication, and had no known drug allergies. The patient also complained of loose teeth and needs replacement of them. The pain was severe, continuous, and aggravated by mastication. No history of trauma revealed. Initially, the swelling was smaller in size, but gradually it increased in size which was not associated with bleeding or discharge. Extraoral examination showed mild swelling with minimal asymmetry on the right-side of the face. The swelling was well-localised in the right maxillary buccal vestibule, extending anteriorly to the right lateral incisor region, posteriorly to the first premolar region, superiorly up to the mucobuccal fold, and inferiorly to the attached gingiva of the involved teeth. Clinically, intraoral examination revealed retained deciduous tooth 53 present in the right maxillary anterior region. A sinus tract opening noted in relation to 12 on the labial gingiva and surrounding mucosa appears erythematous [Table/Fig-1]. On palpation, a localised swelling was firm to hard, non tender, non fluctuant, and non compressible upon palpation.

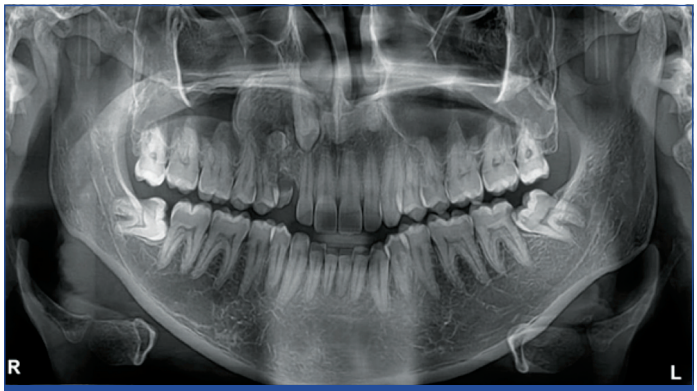
The patient was informed about the nature of the condition, proposed treatment plan, possible risks, benefits, and alternatives and written informed consent was obtained from the patient for both treatment and publication of the images.

An Orthopantomogram (OPG) revealed a well-defined mixed radiopacity and radiolucency extending from 12 to 15 tooth region impacted canine in 13, retained deciduous in 53 and root resorption in 12 and 14 region [Table/Fig-2].

Based on clinical and radiographic examination, a provisional diagnosis of odontogenic tumour was made. Differential diagnosis include odontome, adenomatoid odontogenic tumour, calcifying



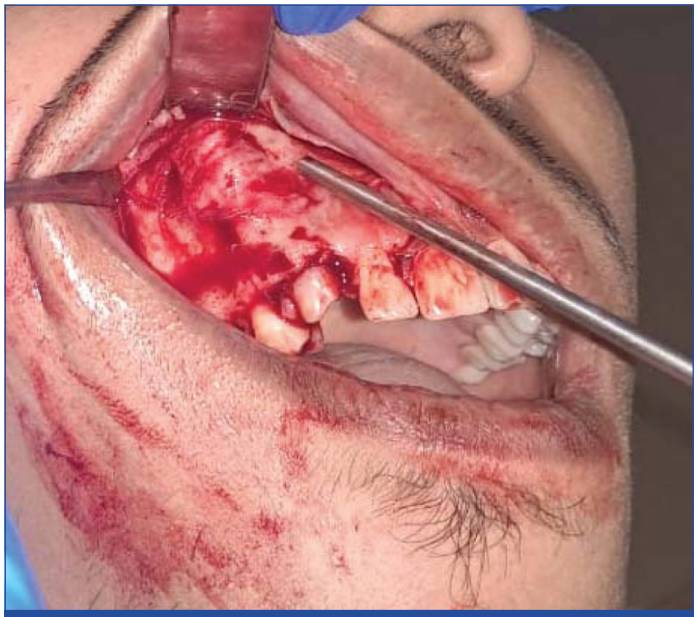
[Table/Fig-1]: Intraoral examination shows swelling in right buccal vestibule near retained deciduous teeth 53 and sinus tract opening are also seen in 12 region.



[Table/Fig-2]: Orthopantomograph shows well-defined mixed radiopacity and radiolucency extending from 12 to 15 tooth region, impacted canine in 13, retained deciduous in 53 and root resorption in 12 and 14 region.

odontogenic tumour, central giant cell granuloma and OF, Some of the lesions will be clinically appear same, the ruling out was done using histopathology. An incisional biopsy was not performed because the lesion was well-defined and of manageable size, allowing complete surgical excision in a single procedure Surgical excision was planned after obtaining patient consent form for surgery, all pre-surgical procedure were followed. Routine blood investigations include bleeding time, clotting time, haemoglobin level and blood sugar level are taken which was apparently normal. Pre-procedural mouth rinse with chlorhexidine given.

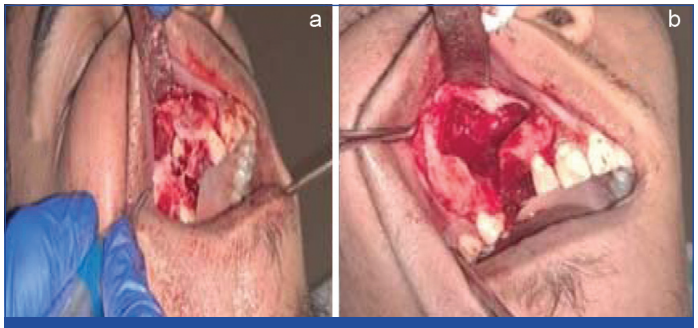
Initially, the surgical area was anaesthetised with 2% lignocaine solution. Bard Parker blade No. 15 was used to give two vertical relieving incision and full thickness mucoperiosteal flap was elevated using periosteal elevator distal to 12 and mesial to 14. These two incisions were connected horizontally by a mid-crestal incision [Table/Fig-3].



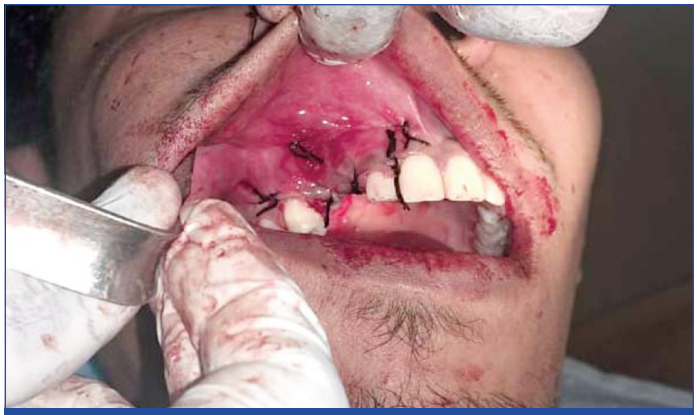
[Table/Fig-3]: Crestal incision and reflection.

Considering the lesion's small size, well-defined margins, and non aggressive radiographic features, excisional biopsy was planned as it would serve both diagnostic and therapeutic purposes. An excisional biopsy was done and the surgical site was curetted [Table/Fig-4a] and irrigated with betadine solution [Table/Fig-4b]. Sterile gauze pressure applied to the surgical site is the most basic and effective method to achieve hemostasis. Care was taken to perform peripheral curettage of the surrounding bone to minimise the possibility of recurrence. The elevated flap was closed and secured with 3-0 simple interrupted non absorbable silk sutures [Table/Fig-5].

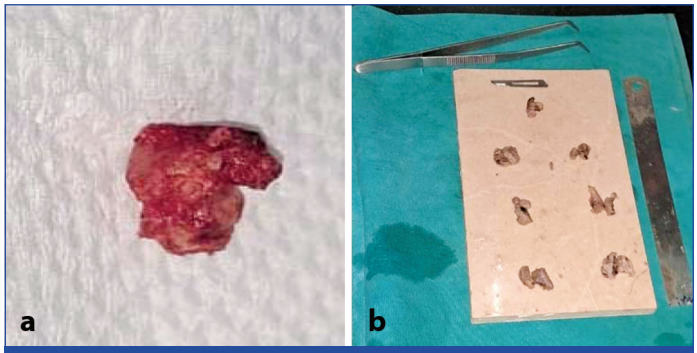
The biopsy specimen included both hard and soft-tissues. It was a mixed type consisting of retained deciduous tooth (53), impacted canine (13) and soft-tissues, which were 3x4 cm (multiple fragments) greyish white, firm to hard in consistency [Table/Fig-6a,b]. These biopsied tissues were preserved in a 10% formalin solution and



[Table/Fig-4]: a} Wound debridement done with surgical bur and bone curettes and Gracey curettes {b) Irrigation with povidone-iodine (Betadine) solution.



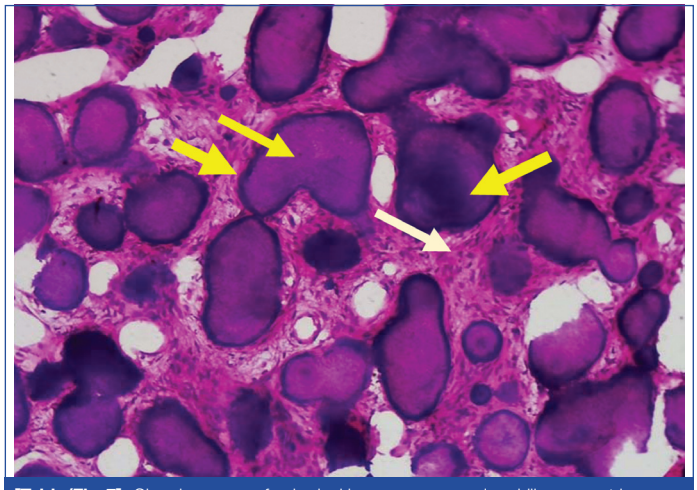
[Table/Fig-5]: Elevated flap was closed and secured with 3-0 silk sutures.



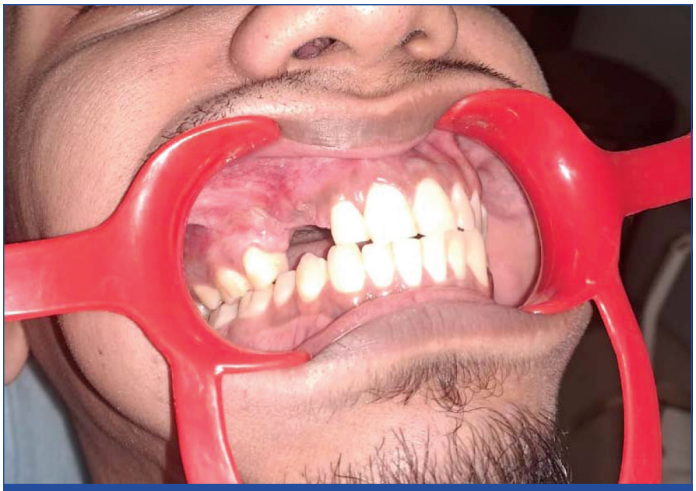
[Table/Fig-6]: Excisional biopsy specimen.

the specimen was sent to the Department of oral and maxillofacial pathology for further histopathological processing.

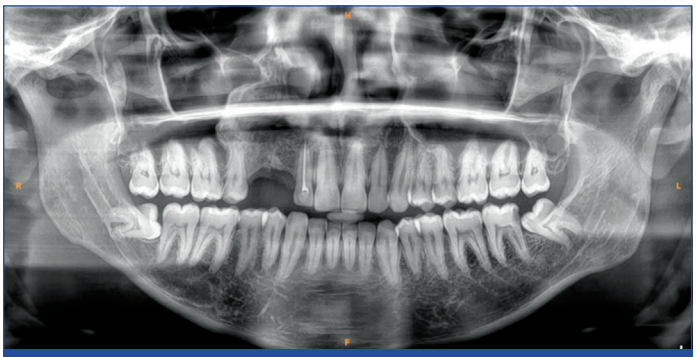
The Haematoxylin and Eosin (H&E) stain revealed a tumour mass composed of fibrous connective tissue. Stellate, spindle-shaped and angulated forms of mesenchymal cell were obtained throughout the lesion. Numerous spherical calcified masses were seen with basophilic central area are surrounded by eosinophilic layer (osteoid). Osteoblastic rimming was observed in many areas [Table/Fig-7]. Focal areas of bony trabeculae were also evident. Based on these features, a diagnosis of juvenile POF was made. The patient was discharged after postsurgical procedure and is under regular follow-up. The postoperative review of the site was after 10 days. The patient was consistently followed-up [Table/Fig-8], and the healing was uneventful without recurrence [Table/Fig-9]. The patient was monitored for a period of three years following surgical excision to evaluate healing and detect any signs of recurrence, post-restoration after one year [Table/Fig-10]. Follow-up visits were scheduled at two-week intervals during the initial healing phase, then monthly for the first six months, and subsequently every six months as part of supportive periodontal therapy.



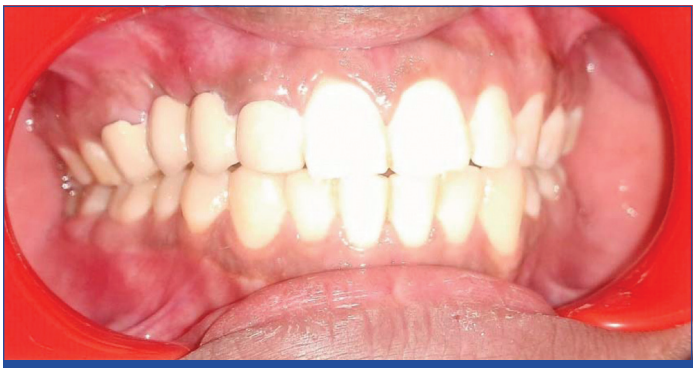
[Table/Fig-7]: Showing areas of spherical homogenous eosinophilic concentric lamella like psammomatoid bodies (40x) surrounded with spindle and stellate-shaped mesenchymal cells (white arrow marking) (H&E, 40x).



[Table/Fig-8]: Postoperative review.



[Table/Fig-9]: Orthopantograph showing postoperative review.



[Table/Fig-10]: Prosthetic restoration after one year.

DISCUSSION

Fibro-osseous Lesions (FOL) are a specific category of lesions affecting the jaws and craniofacial bones [1]. In 2017, the World Health Organisation (WHO) added the term "Fibro-osseous lesion" (FOL) to its classification of odontogenic and maxillofacial bone tumours [2]. OF, classified under FOL, are neoplasms composed of fibrous tissue with a mix of bony trabeculae and cementum-like spherules which differed from the POF. JOF, a type of OF, is diagnosed in early childhood where it has not common in adulthood, with 79% of patients diagnosed before the age of 15 years [3]. The recurrence rate for this lesion is about 30-60% [4].

The POF is a rare subtype of aggressive OF. The most common lesion sites were the paranasal sinuses and the jaw (70% in the paranasal sinuses, followed by 20% in the maxilla and 10% in the mandible), and just 10% occurred in the calvaria [5]. Different terminologies are used in literature as Osteoid fibroma with atypical ossification, POF, JOF, Psammous desmo-osteoblastoma, PsJOF, JOF with psammoma-like ossicles,"Osteoid fibroma with atypical calcification" and "Aggressive of or Active ossifying fibroma" by Varshney A et al., [6].

It commonly affects children and young adults [7], unlike the case reported by Varshney A et al. [6], where the patient was an elderly female aged 50 years [6]. In the literature, even though the age ranges from three months to 72 years, while gradual enlargement has been reported Bohn OL et al., 2011 [8]. In contrast, Slootweg PJ et al., in their study of 33 cases, observed a different distribution pattern, where the majority of lesions were found in the maxilla (6 cases), followed by the paranasal sinuses (2 cases) and the mandible (16 case), with the mandibular lesion occurring in the posterior region [9]. The most common characteristics of patients with PJOF are: ocular proptosis, nasal obstruction, headache, oedema, and epistaxis [10]. In contrast, the present case demonstrated a relatively localised intraoral presentation without associated orbital or sinonasal symptoms. The lesion was identified primarily as a well-defined radiopaque mass in the 12-15 region with no evidence of facial deformity, proptosis, or sinus involvement. This comparatively asymptomatic and localised presentation highlights the importance of early radiographic detection, which can facilitate timely surgical management and prevent the development of more aggressive craniofacial complications.

Imaging modalities like Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) are useful in determining the extent and invasion of tumours. The histopathological variants of JOF include Juvenile Trabecular Ossifying Fibroma (JTOF) and Juvenile Psammomatoid Ossifying Fibroma (JPOF) [11]. Histopathologically, shows calcifications have psammoma-like concentric lamellae with basophilic cores and an eosinophilic osteoid rim. The surrounding fibrocellular connective tissue stroma contains a dense proliferation of spindle-shaped cells with hyperchromatic nuclei [12].

A diagnostic hint for distinguishing these two lesions is that Fibrous dysplasia revealed homogenous radio-dense opacities with a ground-glass appearance that merges into the surrounding normal bone, whereas OF revealed a unilocular mixed radiolucent and radioopaque lesion with strongly defined boundaries [13].

Differentiating between FD lesions and JOF is crucial since the treatment procedures are completely different. JOF, while benign, is enucleated since it has the potential to recur, whereas fibrous dysplasia does not require treatment unless there is functional or cosmetic damage [14].

As previously stated, in present case, the radiologic characteristics revealed a well-defined radio-opacity extending from the 12-15 area, paving the way for a clear diagnosis of PsJOF and removal of the same. The present case report gives a clear differential diagnosis of similar radio opacity in the maxillary anterior region. histopathologic features are imperative for the diagnosis of this rare variant and the aggressive nature should be considered in the treatment plan. Follow up of the patient is necessary for fulfilment of the treatment and complete resolution of the problem.

CONCLUSION(S)

Psammomatoid bodies are the hallmark of histopathological examination for POF. CT scan is necessary for proper diagnosis and treatment plan of the lesion. POF is frequently confused with other neoplasms based on clinical examination and radiographic findings. It is therefore critical to consider this tumour when evaluating osseous head and neck region in younger individuals. Early detection and complete surgical excision followed by long-term follow-up bears importance in clinical management due to their aggressive nature and high recurrence rate.

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PARTICULARS OF CONTRIBUTORS:
1. Professor, Department of Periodontics, Sri Venkateswara Dental College and Hospital, Chennai, Tamil Nadu, India.
2. Professor and Head, Department of Oral Pathology, Sri Venkateswara Dental College and Hospital, Chennai, Tamil Nadu, India.
3. Assistant Professor, Department of Oral Pathology, Sri Venkateswara Dental College and Hospital, Chennai, Tamil Nadu, India.
4. Associate Professor, Department of Periodontics, Sri Venkateswara Dental College and Hospital, Chennai, Tamil Nadu, India.
5. Assistant Professor, Department of Periodontics, Sri Venkateswara Dental College and Hospital, Chennai, Tamil Nadu, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Rathinasamy Kadhiresan,
177/72, Sri Rangammal Street, Old Washermanpet, Chennai-600021,
Tamil Nadu, India.
E-mail: drkadhiresan@gmail.com

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