

A Rare Case of Peripheral Ossifying Fibroma in an Infant

Abstract

Peripheral ossifying fibroma (POF) associated with natal/neonatal teeth is extremely rare. In general, POF occurs as a soft-tissue gingival nodule occurring in young adults. We report an unusual and a rare case in a 5-month-old male child who had two, localized, soft-tissue growth over the mandibular anterior alveolar ridge. History revealed the presence of natal teeth in the same mandibular anterior region, which exfoliated at the age of 2 months. Intraoral periapical radiograph showed soft tissue density with evidence of calcifications that also corroborated with the histopathological finding of masses of mineralized areas. Excision of the lesions followed by histopathological examination proved the final diagnosis of POF. In spite of being a benign reactive lesion, a high recurrence rate has been reported. Such lesions require long-term follow-up subsequent to excision with histopathological examination due to a high recurrence rate.

Keywords: Calcifications, infant, peripheral ossifying fibroma

Introduction

In infants and neonates, the most common localized soft-tissue lesions associated with alveolar ridge are eruption cyst, gingival cyst of newborn, and congenital epulis. Scientific evidence have documented that any kind of localized chronic irritants to gingiva can lead to the development of varied gingival “Reactive lesions.” These “Reactive lesions” generally appear as localized gingival soft-tissue growth and are routinely referred to as localized hyperplastic reactive lesions (LHRLs). The lesion classified under LHRLs is focal fibrous hyperplasia (FFH), pyogenic granuloma (PG), peripheral ossifying fibroma (POF), and peripheral giant cell granuloma.^[1] These lesions are not an uncommon finding in children but extremely rare to occur in infants and neonates. The prevalence of natal and neonatal teeth has been recorded as 1:800 to 1:3000 in different populations with natal teeth being three times more common than neonatal teeth. LHRLs of gingiva in neonates and within the first 10 months of life are very rare, with only 5 cases reported till date [Table 1].^[1] POF associated with the neonatal tooth is even rarer, with only one case reported in the literature.

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We report an extremely rare case of POF involving mandibular anterior alveolar crest associated with natal teeth in a 5-month-old infant having radiographic evidence of calcifications and proven histopathologically. However, this case is unique as no case has been reported so far, which mentions about radiographic evidence of calcifications and associated with the previous history of neonatal teeth.

Case Report

A 5-month-old male child was brought to the Department of Oral Medicine and Radiology with Complaint of swelling in the lower front teeth region, causing difficulty in nursing for the past 3 months. There was a history of two natal teeth in the mandibular incisor region, which exfoliated by itself 3 months back. Subsequently, two small swellings developed at that site, which gradually increased to present size, causing discomfort in nursing. On examination, there were two distinct, firm, soft-tissue pedunculated growth of size 1.5 cm × 1 cm with normal-appearing overlying mucosa in color and texture in relation to the alveolar crest of mandibular central incisor region [Figure 1a]. Intraoral periapical radiograph showed soft-tissue shadow along with irregular radiopacity within it suggestive of calcification or poorly formed teeth and also evidence of deciduous tooth

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Table 1: Review of reported cases of peripheral ossifying fibroma in neonates and infants

Author	Year	Age	Sex	Associated natal/ neonatal tooth	Site	Any radiographic findings reported
Tewari <i>et al.</i> ^[1]	2017	2 months	Male	Neonatal teeth	Anterior mandibular region	No
Cuisia and Brannon ^[2]	2001	6 months	Not mentioned	Not mentioned	Anterior mandibular region	No
Kohli <i>et al.</i> ^[3]	1998	2 h	Female	Neonatal teeth	Anterior mandibular region	No
Buchner and Hansen ^[4]	1987	7 months	Not mentioned	Not mentioned	Not mentioned	No
Yip and Yeow ^[5]	1973	7 days	Female	Not associated	Right Maxillary Molar region	No

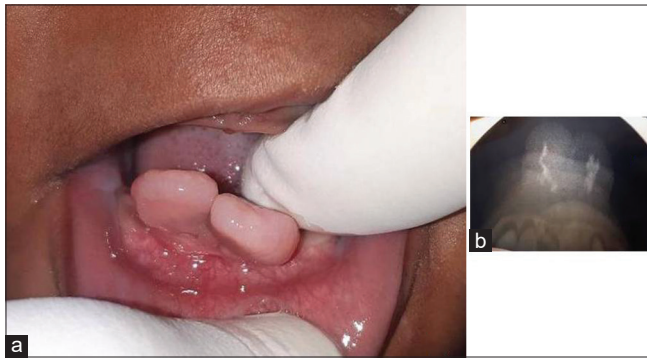


Figure 1: Intraoral clinical photograph showing two distinct, pedunculated, firm, soft tissue growth attached to alveolar crest of mandibular anterior region (a) and intraoral periapical radiograph of mandibular anterior region showing soft tissue shadow along with irregular radiopacity within it suggestive of calcification and underlying tooth buds of primary teeth (b)

buds present below the alveolar crest [Figure 1b]. Based on the history of natal teeth, clinical findings and radiographic evidence of soft tissue density lesion with calcifications, a provisional diagnosis of FFH with calcifications was made. It was decided to excise the lesion, and informed consent was taken from the parents. Routine blood investigations, including complete hemogram, total and differential leukocyte count was performed, all of which were within the normal limits. Under local anesthesia, the excision of the lesion was performed in toto using the blade. During excision, minor hemorrhage was controlled using pressure packs. The mucosa overlying the ridge was sutured with silk suture. The tissue was sent for histopathologic examination. The patient was recalled after 1 week for suture removal, and post-operative healing was uneventful [Figure 2a]. Postoperative intraoral radiograph was taken, which revealed normal alveolar mucosa with tooth buds of anterior teeth [Figure 2b]. On gross examination of the excised tissue [Figure 3]; no remnants of natal teeth were found. Histopathology revealed parakeratinized hyperplastic stratified squamous epithelium having long and narrow rete ridges. Superficial connective tissue was consisting of irregularly arranged moderate collagen bundles, fibroblast, and varying sized blood vessels with diffuse mild lymphocytic infiltration. Deeper connective tissue comprised of hypercellular areas having plump fibroblasts with vesicular nuclei and also varying degrees of mineralized areas in the form of irregular large masses

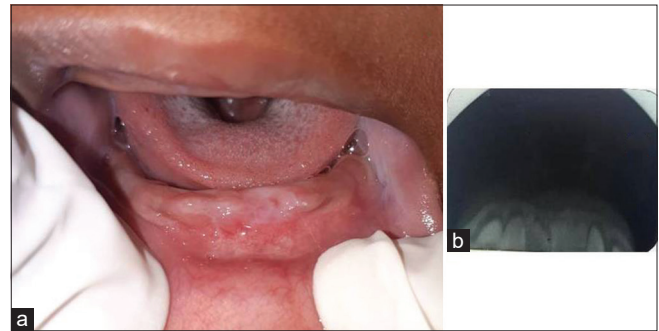


Figure 2: Intraoral clinical photograph showing 2 weeks post-operative photograph with uneventful healing (a) and postoperative intraoral periapical radiograph of mandibular anterior region showing normal alveolar ridge with underlying tooth buds of primary teeth (b)

was seen. A final histopathological diagnosis of POF was made [Figure 4a and b]. The patient is under follow-up.

Discussion

POF, also known by other synonyms such as peripheral fibroma with calcification, ossifying fibrous epulis, the calcifying fibroblastic granuloma is one of the benign, reactive, proliferative lesions exclusively involving gingival mucosa. It is most likely to be originating from the cells of periodontal ligament (PDL) according to the various hypothesis proposed in the literature and trauma, local irritants such as calculus, plaque, ill-fitting restoration, and appliances have been attributed toward its etiology in the form of chronic irritants.^[2] Therefore in the present case, presence of natal teeth might have been the source of chronic irritation leading to the development of POF.

POF is frequently prevalent in the second to third decade of life with female predilection and clinically appears as solitary, red to pink, pedunculated or sessile, nodular, soft-tissue growth commonly seen in gingival interdental papilla area of maxillary anterior region.^[1] However, the reported prevalence in the age group of 0–10 years has been 1%–2% only and POF associated with natal and neonatal teeth in neonates and infants has been extremely rare entity with only 5 cases reported till date [Table 1].^[1-5]

The radiographic appearance is variable with evidence of soft-tissue shadow, demonstrating nil to the varying degree of calcifications, and sometimes, larger lesions also reveal erosion of underlying alveolar bone and may

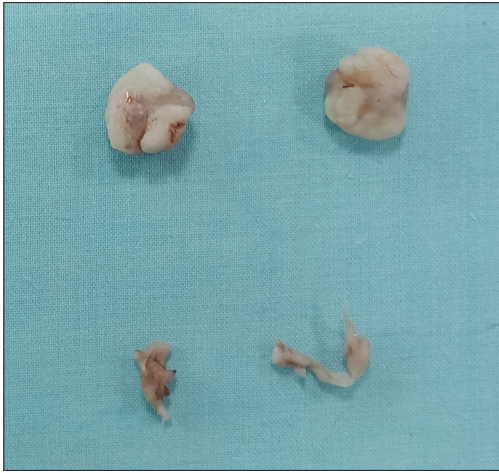


Figure 3: Gross picture of the excised specimen

also cause displacement of adjacent teeth. The presence of radiographic finding of calcifications, which is commonly seen in adults, was in contrast to the present case. The peculiar histopathological features include dense fibrocellular proliferation with random focal deposits of calcified material varying from irregular dystrophic/metaplastic calcification to laminated concentric deposits resembling Liesegang ring.^[1] The degree of mineralized deposits is related to the maturation of the POF lesion.

Treatment commonly rendered is surgical excision of the pathology with curettage of associated periosteum and removal of local irritants to prevent the recurrence. However, in children, treatment should be more aggressive ensuring deep excision as these lesions display an exuberant growth rate and may also cause erosion of underlying bone, displacement of teeth as well as interfere with the eruption of teeth.^[2] With the advent of lasers in dentistry, recently, lasers have been employed for excision of POF. Lasers offer many advantages over conventional surgery, including scalpel such as bloodless surgical site, reduced trauma with minimal scarring, reduced chances of infection, and uneventful healing.^[6] Some authors have also advocated the use of Piezosurgery that allows for enhanced visibility of the surgical site, reduced bleeding, and less discomfort to the patient.^[7] A high recurrence rate of 16%–20% and ambiguous clinical picture necessitates a histopathological examination after their excision, and a long-term follow-up.^[8,9]

Conclusion

- This case report warrants a close follow up of patients with natal/neonatal teeth since they could be a source of irritation that may lead to localized gingival soft tissue reactive lesions
- POF should also be considered in the differential diagnosis of localized soft tissue gingival growth, especially when there is evidence of focal calcifications seen on the radiograph
- Patients with POF should be kept under vigilant long-term follow-up, particularly when diagnosed in children.

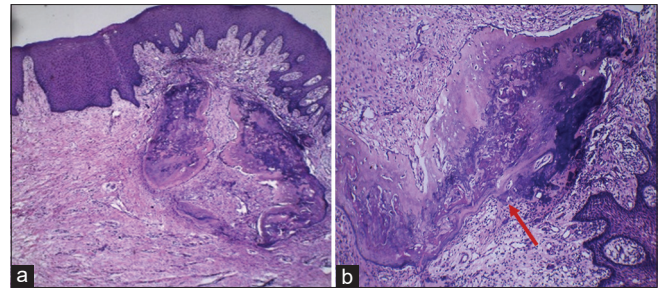


Figure 4: Microphotograph (H and E, ×4) showing parakeratinised hyperplastic stratified squamous epithelium having long and narrow rete ridges, connective tissue was consisting of collagen bundles, fibroblast, diffuse mild lymphocytic infiltration and mineralized areas (a) and Microphotograph (H and E, ×40) showing varying degree of mineralized areas in connective tissue shown by an arrow and hypercellular areas having plump fibroblasts (b)

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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