



CASE REPORT

PERIPHERAL OSSIFYING FIBROMA AND GINGIVAL PIGMENTATION: A CASE REPORT

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ABSTRACT

Peripheral ossifying fibroma is a reactive lesion of the gingival tissues, usually originates from the periodontal ligament cells. It is characterized by highly cellular connective tissue exhibiting dystrophic calcification with the formation of bone and cementum-like materials. It predominantly occurs in females compared to that of males and occurs in relation to maxillary teeth particularly the anterior to the molars. A 14 years old girl child was reported with a localised gingival overgrowth in relation to the 11, 21 and 22. The growth was excised and healing was found uneventful. However, removal of the growth revealed the unesthetic black colored gingiva in adjacent areas in contrast of normal colored (coral pink) gingiva at the surgical area. It subsequently demands the depigmentation of gingiva, which was carried out using scalpel. The present case was under observation for considerably a long time and shows no sign of recurrence.

INTRODUCTION

Localised gingival overgrowth, specifically the reactive lesion, is regarded as 'epulis'. Reactive lesions comprises a group of fibrous connective tissue lesions that commonly occur in the oral mucosa as a result of injury. Various categories of localized gingival overgrowth are identified, namely pyogenic granuloma (including pregnancy tumor), peripheral ossifying fibroma (ossifying fibroid epulis, peripheral fibroma with calcification, calcifying fibroblastic granuloma, and peripheral odontogenic ossifying fibroma), peripheral fibroma (fibrous hyperplasia) and Peripheral giant cell granuloma (Neville *et al.*, 2009). Amongst these, peripheral fibroma is the most prevalent one (56 - 61%) followed by pyogenic granuloma (19 - 27%), peripheral ossifying fibroma (10 - 18%), and peripheral giant cell granuloma (1.5 - 7%) (Zhang *et al.*, 2007; Kfir *et al.*, 1980). Peripheral ossifying fibroma occurs exclusively on gingiva. It usually arises from the interdental papillae and is thought to originate from the cells of the periodontal ligament (Kumar *et al.*, 2006). It represents upto 2% of all the oral lesions that are biopsied and can occur at any age although more common in children and young adults with the peak incidence at 13 years. It most often occurs in the maxilla, particularly anterior to molars and predominantly affects the women (Shafer *et al.*, 1983) suggesting that role of hormone in its etiology (Effiom *et al.*, 2011). Clinically, peripheral ossifying fibroma appears as a pink to red coloured

nodular mass, either pedunculated or sessile. The surface of the mass may be either smooth or ulcerated. Migration of teeth with interdental bone destruction has been reported in some cases. The definitive diagnosis is based on histological examination with the identification of a highly cellular connective tissue and focal presence of bone, cementum, or irregular amounts of dystrophic calcification. Surgical excision including the periodontal ligament and periosteum at the base of the lesion is the preferred treatment modality in order to reduce the chances of recurrence (*i.e.* 8% to 20%) (Eversole and Rovin, 1972).

CASE REPORT

A 14 year old girl reported with the chief complaint of black gums and progressive painless growth in relation to 11, 21 and 22, which started as a small swelling approximately 9 months ago and gradually increased. Intraoral examination revealed a localised pale pink, sessile gingival overgrowth in relation to the maxillary incisors measuring approximately 2 cm x 1.5 cm in size, extending from the distal aspect of 11 to the mesial aspect of 22. The growth was extended coronally upto the incisal edges of 11, 21 and 22, and towards palate (Figure 1A). The growth was blanched on digital pressure. The lesion was nontranslucent, noncompressible, nonpulsatile, not fixed to underlying tissues and revealed no blood on aspiration. There was no history of trauma or injury to that area. The teeth in relation to the growth were vital and not mobile.

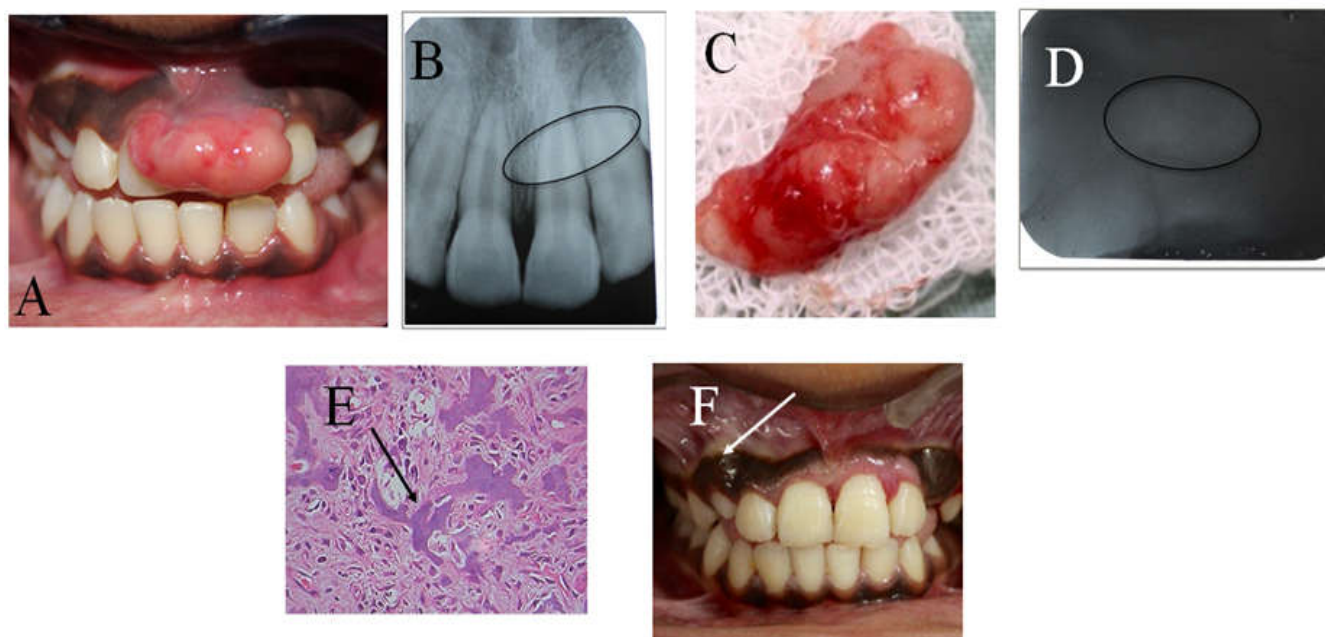


Figure 1. Peripheral ossifying fibroma. (A) Intraoral facial view of pale, pink and firm gingival overgrowth, extending from distal aspect of 11 to mesial aspect of 22, covering the entire crown surface labially extending up to the incisal edges of 11, 12 and 21. (B) IOPA x-ray reveals radiopaque shadow of the growth superimposed over the roots of 21 and 22. (C) Excised lesion, approximately of 2 x 1.5 cm in size. (D) IOPA x-ray of the excised tissue showing radiopacity. (E) Histologic picture. H&E staining showing Connective tissue revealing foci of metaplastic bone rimmed with osteoblast and numerous acellular dark staining calcified tissues, confirming the diagnosis of peripheral ossifying fibroma. Original magnification (x 40). (F) Postoperative facial view after one month

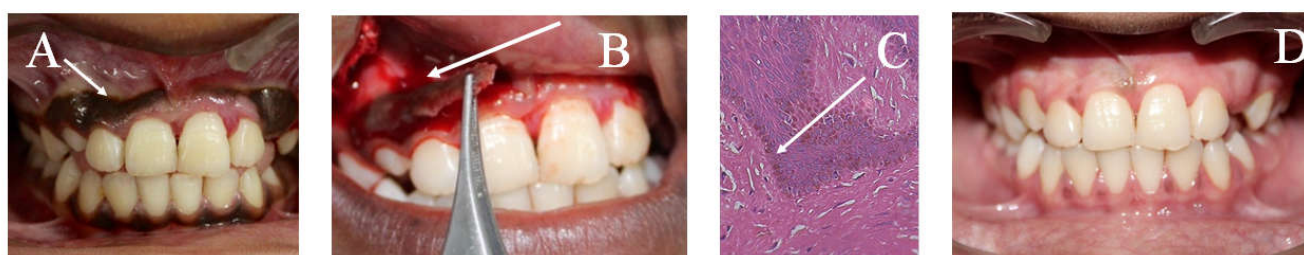


Figure 2. (A) Black-colored gingival pigmentation, shown by arrow. (B) Depigmentation carrying out using scalpel by surgical slicing technique. (C) Histologic picture. H&E staining shows numerous dark stained melanocytes in the basal layer of pigmented gingival epithelium (D) Postoperative facial view after six month follow-up. Note that there is no sign of recurrence of both the gingival pigmentation and growth

Oral hygiene was fair, but gingiva was observed to be a continuous band of black colored. Gingival discoloration may be physiologic (*e.g.* racial pigmentation) or pathologic, which is usually seen as manifestation of certain systemic diseases, such as in Addison's disease and in malignant neoplasms, namely melanoma and Kaposi's sarcoma (Kauzman *et al.*, 2004). Physiologic gingival pigmentation usually results from melanin pigment, a non-hemoglobin-derived brown pigment produced by melanoblasts present in the basal and spinous layers of the gingival epithelium (Thangavelu *et al.*, 2012). Melanin pigmentation clinically manifests as a diffuse deep purplish discoloration or as irregularly shaped brown or black patches or strands. Gingival pigmentation is asymptomatic and does not cause any medical problems. However, it may be an esthetic concern for the patient and demands its removal, referred to as gingival depigmentation (Deepak *et al.*, 2005). An intraoral periapical radiograph of the region revealed a radiopaque shadow corresponding to the location of the growth, suggestive of calcified lesion (Figure 1B). Extra orally, there was no swelling and the overlying skin was normal in color with no elevation of temperature. However, the upper lip was observed to be incompetent as lip approximation was difficult due to the growth.

Lymph nodes were non palpable. Family history was noncontributory. The patient had no relevant systemic history. Complete haemogram of the patient was found to be within normal limits. The treatment plan included Phase I therapy, comprising of scaling and root planing, followed by surgical excision. Consent for the surgical procedure was obtained from the patient. The lesion was excised along with the surrounding normal tissue using Kirkland's Gingivectomy knife under local anaesthesia. After achieving adequate haemostasis, periodontal dressing was placed. Antibiotics and analgesics were prescribed. The patient was recalled after 7 days. The tissue was sent for histopathologic examination. Periodontal dressing was removed after a week, the surgical site showed signs of healing.

Based on the clinical examination, following were considered for differential diagnosis

Peripheral fibroma: It occurs as a firm, round to oval, smooth surfaced, pale pink colored raised mass with either pedunculated or sessile base. It is more common in anterior maxilla, common in fourth to sixth decade of life and shows female predilection (Kfir *et al.*, 1980).

Peripheral giant cell granuloma: It appears as a deep red to reddish blue, raised lesion with surface ulceration, which is either sessile or pedunculated. It commonly occurs in fourth to sixth decade of life and shows predilection to males. It is more common in mandible than maxilla (Binnie, 1999).

Pyogenic granuloma: It is an exuberant tissue response to local irritation or trauma. It is highly vascular, red or reddish purple, often elevated and ulcerated, and bleeds easily. It is also believed that this lesion may develop into a peripheral ossifying fibroma or peripheral fibroma over time through fibrous maturation and ossification (Binnie, 1999). In the reported case, the microscopic examination of the excised tissue revealed acanthotic stratified squamous epithelium with elongated and branched rete pegs. Connective tissue showed dilated vascular channels with foci of metaplastic bone rimmed with osteoblast, numerous acellular dark staining calcified tissues and collections of lymphoplasmic cells and neutrophils suggestive of a reactive lesion with calcification. (Figure 1F). Considering the clinical and histopathological findings, a final diagnosis of peripheral ossifying fibroma was established. The follow up on day 30, the newly healed wound area was found to be normal coral pink in contrast of the adjacent pigmented areas (Figure 1 C) and the patient was very disagreeable to this black colored gingiva (Figure 2A). Considering the patient's concern, gingival depigmentation was planned. Accordingly, a slice containing entire pigmented epithelium along with a thin layer of connective tissue was removed using a #15 blade (Figure 2B). Then the wound was irrigated with normal saline. Pressure was applied with sterile gauze to stop the bleeding, followed by the application of the Coe pack as periodontal dressing for a week. The excised gingival tissue revealed numerous dark stained melanocytes in the basal layer of gingival epithelium on histological evaluation (Figure 2C). The healing was found to be uneventful without any postsurgical complications. The gingiva appeared firm in consistency and pink in color with no occurrence of pigmentation up to 6 months of surgical observation (Figure 2D).

DISCUSSION

Peripheral ossifying fibroma is considered as a reactive hyperplastic lesion of gingiva. Shepherd first reported this entity as "alveolar exostosis" in 1844 but the term peripheral ossifying fibroma was coined by Eversole and Rovin in 1972. Later on, Bhaskar *et al.*, in 1984 described this lesion as peripheral fibroma with calcification. Different terminologies, such as fibrous epulis, calcifying fibroblastic granuloma or peripheral fibroma with calcification have been used in the literature to describe it (Bhaskar and Jacoway, 1966). Intraoral ossifying fibromas have been described in the literature since the late 1940s. Two types of ossifying fibromas are identified, namely central and peripheral. The central ossifying fibroma arises from the endosteum and/or the periodontal ligament, adjacent to the root apex and expands from the medullary cavity of the bone, while peripheral type occurs on the soft tissues overlying the alveolar process (Miller *et al.*, 1990). In addition, peripheral ossifying fibroma is associated with some known genetic mutations, referred to as multicentric seen in nevoid basal cell carcinoma syndrome, multiple endocrine neoplasia type II, neurofibromatosis and Gardner syndrome (Kumar *et al.*, 2006). In initial stage, peripheral ossifying fibroma may not show any detectable amount of mineralization but mature lesion shows flecks and patches of radiopacity in

radiographs (Neville *et al.*, 2009). The radiopacity of teeth structures usually obscure the calcified mass of the lesion in radiographs, so it is prudent to take a radiograph of excised lesion (Jyothi and Rao, 2012). Here, IOPA x-ray revealed a well-defined radiopaque shadow of the lesion superimposing over the roots of maxillary incisors (Figure 1B). Further, radiopacity of the excised lesion in x-ray suggests calcification of the lesion mass (Figure 1D). The exact origin of peripheral ossifying fibroma is not known. However, it is believed that it originates from the periodontal ligament cells considering the fact that it commonly arises from the interdental papilla (which is in close proximity to periodontal ligament). In addition, the oxytalan fibers found within the mineralised matrix of some lesions, may originate from the periodontal ligament. (Eversole and Rovin, 1972; Kumar *et al.*, 2006). The widely accepted pathogenesis of peripheral ossifying fibroma is the inflammatory hyperplasia of the periodontal ligament cells to local irritants such as dental plaque, calculus or tissue trauma, which leads to the metaplasia of the connective tissue resulting in initiation of bone formation or dystrophic calcification (Eversole and Rovin, 1972). Confirmatory diagnosis is based on the histopathological observation of the biopsy specimen. The characteristic feature of the peripheral ossifying fibroma is the presence of highly cellular connective tissue containing foci of calcified material (in form of bone, cementum or osteoid) along with the numerous proliferating fibroblasts, variable number of acute and chronic inflammatory cells, and ulcerated/intact epithelial lining.

The intact epithelial lining observed here indicates a non-ulcerated peripheral ossifying fibroma, which is similar to an ulcerated type except for the presence of intact surface epithelium (Nazareth *et al.*, 2011) (Figure 1E). Local surgical excision including the involved periodontal ligament and periosteum is the ideal treatment of peripheral ossifying fibroma. It recurs due to incomplete removal of the lesion, failure to eliminate local irritants and difficulty in accessing the lesion during surgical manipulation. Since the recurrence rate is high (8 to 20%), it demands long term postoperative monitoring (Eversole and Rovin, 1972). Physiologic pigmentation of gingiva is probably genetically determined. However, Dummett in 1967 suggested that the degree of pigmentation is partially related to mechanical, chemical and physical stimulation. Removal of gingival pigmentation (depigmentation) is demanded for better esthetics. The treatment modalities for depigmentation are scalpel, electro surgery, cryosurgery and lasers. However, selection of the method depends on the clinical experience, patient's affordability and individual preferences. In this case, depigmentation was carried out by scalpel. The healing was seen uneventful with minimum postoperative discomfort to the patient. The initial result of the depigmentation is highly encouraging, repigmentation (reappearance of pigmentation) is a common problem. Though the exact mechanism of repigmentation is not known, it may occur due to migration of the active melanocytes to the treated area from the adjacent pigmented tissues (Doshi *et al.*, 2012). Thus, long term follow-up of these cases is necessary. The present case is still under observation even after seven months of surgical removal of growth and pigmentation and shows no sign of recurrence of both the growth and pigmentation.

CONCLUSION

Peripheral ossifying fibroma is a reactive lesion of gingiva with limited growth potential. Proper diagnosis of these lesions

is essential to treat as well as to prevent the recurrence. Again, gingival pigmentation is an esthetic concern for the patient though it does not cause any medical problem. Since recurrence rate of both peripheral ossifying fibroma and pigmentation are high, thus it demands long term follow-up.

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