Case Report

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Dentistry Section

Atypical Presentation of Lobular Capillary Haemangioma of the Maxillary Alveolus: A Case Report

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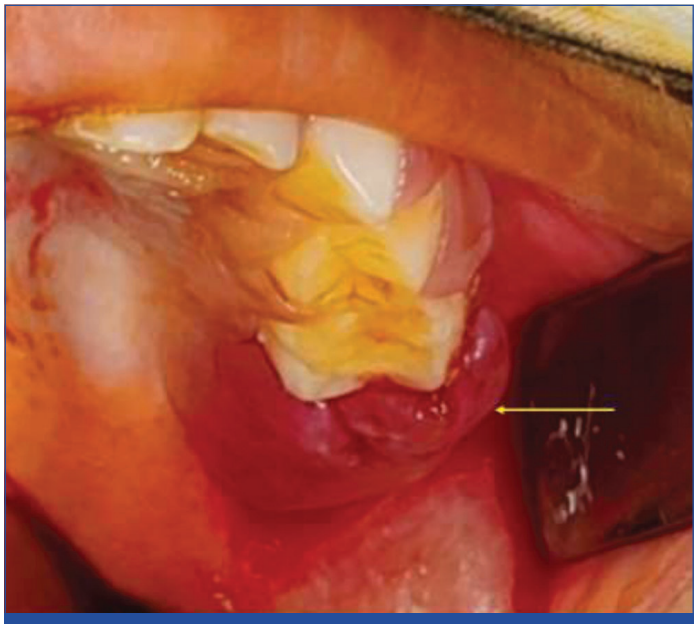
ABSTRACT

Pyogenic Granuloma (PG) is a benign vascular lesion that rapidly grows, is painless, and presents as a friable, smooth, or lobulated exophytic lesion, manifesting as small, red, erythematous papules with a pedunculated or sessile base. The most common site is the gingiva, followed by the tongue and buccal mucosa. Hereby, the authors present a case of a five-year-old girl who reported for an oral and maxillofacial consultation with a swelling in her left upper gum region, associated with multiple episodes of intermittent bleeding. On presentation, a bluish-red, sessile lesion was observed on the gingiva of the left upper 1st molar. Magnetic Resonance Imaging (MRI) and Angiography provided a provisional diagnosis of a vascular tumour, following which embolisation and excision of the lesion were performed by Interventional Radiologists (IR) and Maxillofacial Surgeons. The patient's recovery was uneventful, and histopathology{Haematoxylin and Eosin (H&E)} was reported as Lobular Capillary Haemangioma (LCH).

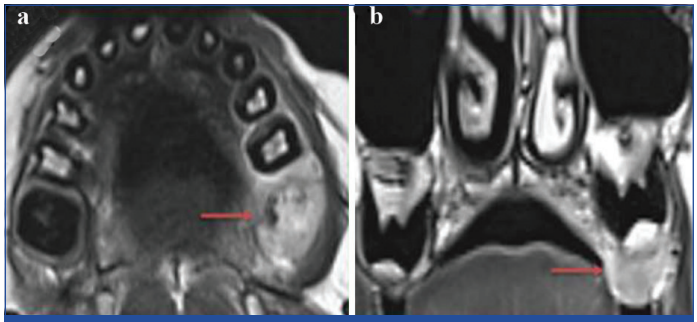
Keywords: Angiography, Neoplasms, Papule, Vascular tissue

CASE REPORT

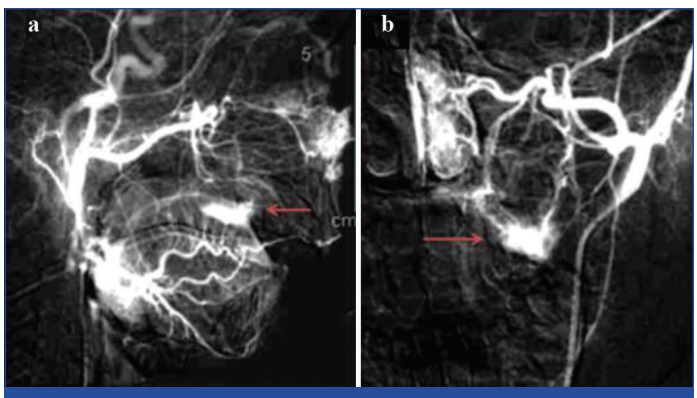
A five-year-old girl reported to the Oral and Maxillofacial Surgery Department at Christian Medical College and Hospital in Vellore, Tamil Nadu, India, with a chief complaint of swollen gums for almost two years, along with intermittent episodes of spontaneous, profuse bleeding that settles on its own. The swelling had been gradually increasing in size. There was no relevant medical or familial history, and no deleterious oral habits were reported. Upon examination, no gross facial asymmetry was present. Intraorally, a 2×2 cm sessile lesion, bluish-red in colour, was visible, arising from the distal aspect of the gingiva of the left maxillary deciduous 2nd molar. The surface of the lesion was smooth, with no ulceration, pulsation, or bruit noted. The lesion started to bleed on palpation and settled with pressure pack placement [Table/Fig-1]. MRI imaging of the face and neck with contrast revealed a 1.5×0.6×1.3 cm {Anterioposterior (AP)×Craniocaudal (CC)×Transverse (TR)} soft-tissue lesion in the upper alveolus, superficial to the molars. The lesion showed intermediate intensity on T1 and T2 and was hyperintense on T2-Short Tau Inversion Recovery (STIR), with homogeneous postcontrast enhancement. There were no intralesional vessels and no deep extension into the alveolus. Features of an Arteriovenous Malformation (AVM) were absent. Radiologically, it was determined that the growth was a vascular tumour. Under local anaesthesia, an angiography was performed. A retrograde right common femoral artery access was used to perform the angiography. Focal blush was seen in the superior alveolar arch on the left-side, superficial to the molar teeth, with multiple small arterial feeders from alveolar branches of the left internal maxillary artery [Table/Fig-2a,b-3a,b]. Based on the clinical presentation, it was provisionally diagnosed as an Arteriovenous Malformation (AVM), and with radiological investigations, it was



[Table/Fig-1]: Intraoral photograph showing the lesion in left posterior maxilla distal to the last molar present.

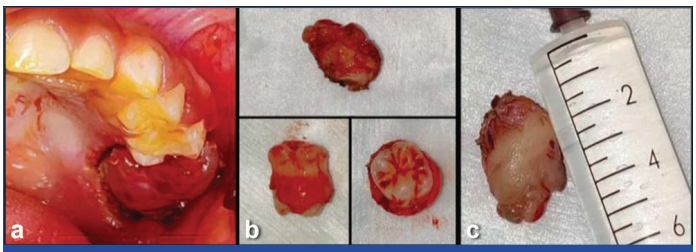


[Table/Fig-2a-b]: Magnetic Resonance Imaging (MRI) face with contrast showing the lesion in the left maxillary alveolus with a tooth above.

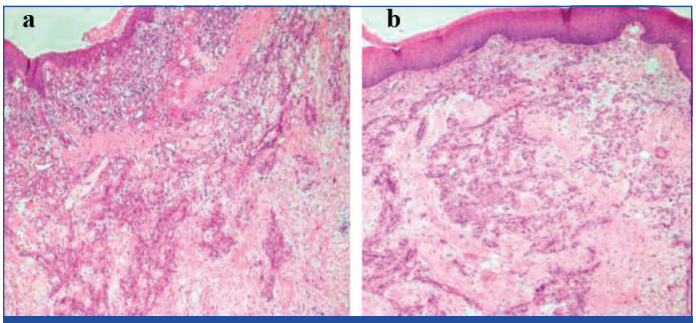


[Table/Fig-3a-b]: Invert image of angiography with blush in left maxillary alveolus with feeders from alveolar branches of internal maxillary artery.

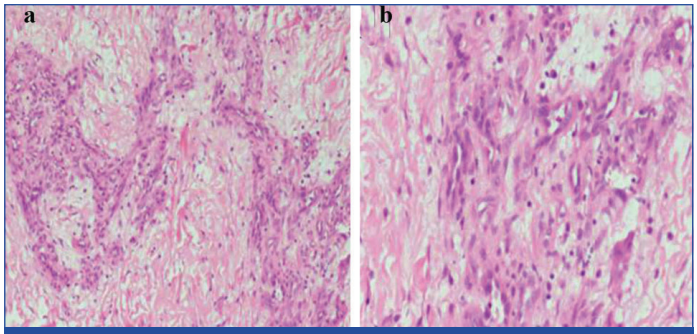
diagnosed as a vascular tumour. Under general anaesthesia, as a first step, embolisation of the lesion was done by Interventional Radiology (IR), followed by excision of the lesion with the extraction of tooth 26. For the embolisation, access was through the right common femoral artery. A 4F short sheath, vertebral glide, and Progreat catheter were used. Cannulation of the left External Carotid Artery (ECA) and the internal maxillary artery was done. A 1000-micron Polyvinyl Alcohol (PVA) particle was the agent used. A pre-embolisation contrast run revealed abnormal contrast blush in the left maxillary gingiva region. Post-embolisation contrast run showed a near complete absence of abnormal blush. From the IR procedure room, the patient was shifted intubated to the maxillofacial operating theatre, and under aseptic precautions, an intra-oral excisional biopsy was performed. During the surgery, it was noted that the lesion was firmly adherent to the underlying 26, thus it was also extracted along with the excision specimen [Table/Fig-4a-c]. Primary closure was done with the advancement of buccal mucosa, and intraoperative blood loss was negligible. Recovery from anaesthesia was uneventful, and the patient was shifted to the ward for postoperative care. Haematoxylin and Eosin (H&E) staining showed polypoidal fragments of stratified squamous epithelium with focal ulceration lined by fibrin and acute inflammatory exudate. The immediate subepithelium shows several proliferated, interconnected, and compressed thin-walled vascular channels lined by plump endothelial cells [Table/Fig-5a,b-6a,b]. The deeper part of the subepithelium shows parts of an ill-circumscribed lesion composed of haphazard and loosely arranged spindle cells displaying mild nuclear pleomorphism, dispersed chromatin with some small nucleoli, and indistinct cytoplasmic borders. Entrapped islands of odontogenic epithelium are seen within the lesion. The stroma, which was alcian blue positive, showed extensive myxoid change. The periphery shows occasional multinucleate giant cells. The features were suggestive of PG (LCH with ulceration) with oral focal mucinosis of the left posterior maxillary alveolus [Table/Fig-7a,b]. Postoperatively, the patient was stable and discharged on the 1st postoperative day on antibiotics and analgesics. A review in the outpatient department after one week was carried out. It was noted that mouth opening was reduced secondary to pain. Wound healing at the procedure site was satisfactory, and there was no evidence of any oro-antral communication. The patient had not reported for three months and six months visits as she did not have any specific complaints. The patient reported at the end of the first postoperative year, and reassessment revealed no evidence of recurrence [Table/Fig-8].



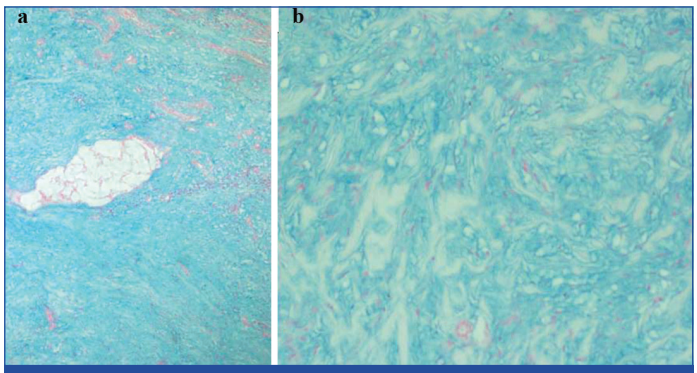
[Table/Fig-4a-c]: Intraoperative excisional biopsy photographs. The excised specimen along with the tooth to which it was attached is seen.



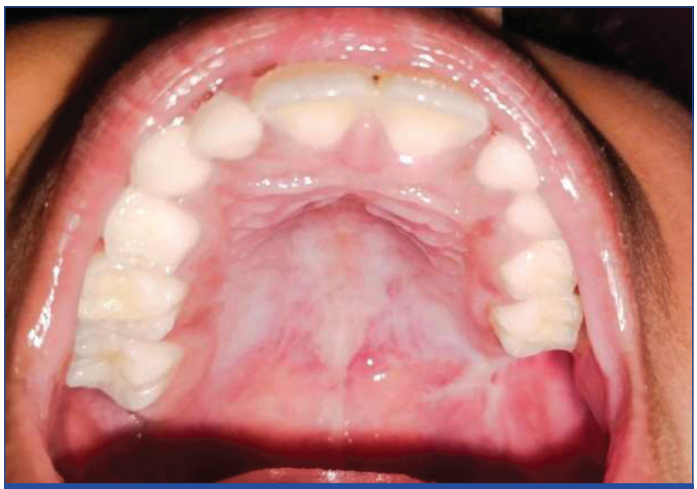
[Table/Fig-5a-b]: Lobular Capillary Haemangioma (LCH), (H&E, 40X).



[Table/Fig-6a-b]: a) Vascular channels lined by plump endothelial cells (H&E,100X); b) Vascular channels lined by plump endothelial cells (H&E, 200X).



[Table/Fig-7a-b]: a) Alcian blue staining- Haemangioma associated with oral focal mucinosis (H&E, 40X); (b) Alcian blue staining demonstrating the myxoid stroma (H&E,100X).



[Table/Fig-8]: Intraoral one year postoperative photograph showing satisfactory healing and no clinical features of recurrence.

DISCUSSION

Pyogenic Granulomas (PG) are commonly occurring reactive growths often seen in the oral cavity, presenting as a response to tissue irritation, trauma, hormonal imbalances, and graft reactions [1]. They are vascular lesions of the skin and mucous membranes. Initially referred to as "Botryomycose Humane," Hartzell in 1904 is credited with giving it the term PG [2]. The term "haemangiomatous granuloma" accurately expresses the histopathologic picture (haemangioma-like) and inflammatory nature (granuloma) of an oral PG [2]. The name "pyogenic" is currently seen as misleading as the condition does not arise from an infection but rather from chronic irritation and the tissue's reaction to that irritation [3,4]. According to the latest International Society for the Study of Vascular Anomalies (ISSVA) classification put forward in 2018, PGs, also known as lobular capillary haemangiomas, are considered benign vascular tumours [5]. PGs are most frequently seen in the head and neck region, with a strong predilection for the oral cavity, particularly the tongue and gingiva [6,7]. Only 15% of the tumours have an alveolar extension, more common in the maxillary anterior than the posterior region [8-10]. The lesion in

present case was present on the left maxillary alveolus posterior to the last deciduous tooth, where there were no obvious local factors causing chronic irritation. Clinically, the majority of PGs are red, smooth, or lobulated with haemorrhagic and compressible characteristics. As a result of subsequent damage, they frequently become ulcerated and covered by a yellow pseudomembranous slough due to subsequent damage [11]. In present case, the lesion was sessile, non proliferative, very vascular, and bled profusely with mild provocation. As it was bleeding on provocation, it was suspected to be an arteriovenous malformation. Consequently, an MRI with contrast of the head and neck was done which reported a small vascular tumour with no features of AVM. Since the lesion was vascular and the clinical diagnosis was inconclusive, embolisation of the lesion followed by surgical excision under general anaesthesia was performed. However, the histopathology revealed it to be a capillary haemangioma. LCH and non LCH are two distinct histopathological types of PG [1]. The lesion in present case was diagnosed as a haemangioma of the left maxillary alveolus based on its clinical presentation and histopathologically as a PG of LCH type. The standard treatment for oral PG includes eliminating the aetiological factors and conservative surgical removal of the lesion. Various surgical modalities have been used to excise oral PG, including cryosurgery, cauterisation with silver nitrate, sclerotherapy, injection of absolute ethanol, sodium tetradecyl sulfate and corticosteroids, as well as laser treatments such as Neodymium-doped Yttrium Aluminum Garnet (Nd:YAG) and CO_2 laser, as well as laser photocoagulation [12-14]. There is insufficient data in the literature to advocate for embolisation for LCH as surgical excision was the treatment of choice. However, since the lesion was radiologically and clinically similar to AVM, a combination therapy involving embolisation and surgical excision was selected. Colour-Doppler ultrasound offers a non invasive, economical, real-time assessment of oral anomalies and is currently regarded as the first-line imaging method. It provides morphological and vascular data helpful in identifying effective treatment alternatives [15]. In cases involving children and pregnant women, choosing an appropriate treatment plan is imperative to limit morbidity and reduce the risk of recurrence [16]. To address a case of PG with an unusual presentation, a combination therapy consisting of embolisation and surgical excision was selected. In the year following surgery, the patient did not exhibit any recurrence symptoms.

CONCLUSION(S)

The LCH, a histological subtype of PG, is a benign vascular tumour for which surgical excision with removal of local irritants is the primary treatment of choice unless the patient is unfit for the surgical procedure. Children with LCH may present with a sessile, non proliferative mass. Treatment of lesions that tend to bleed can have serious medical repercussions if not carried out with sufficient investigation and a precise diagnosis. Accurate investigations and diagnosis are crucial for treating bleeding lesions. Histopathologic analysis is the only way to determine the definitive diagnosis.

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