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RESEARCH ARTICLE

AN UNUSUAL CASE OF MULTIPLE HEMANGIOMAS WITH MULTIPLE PHLEBOLITHS IN ORAL CAVITY

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ABSTRACT

Haemangioma is the most common benign tumour of vascular origin of the head and neck region. The possible sites of occurrence in oral cavity are lips, tongue, buccal mucosa, and palate. Despite its benign origin and behaviour, it is always of clinical importance to the dental profession and requires appropriate management. Phleboliths are calcified thrombi found in veins, venulae and sinusoidal vessels of hemangiomas formed due to Changes in blood flow dynamics in the head and neck. Phleboliths nearly always signal the presence of a hemangioma. This article presents a relatively rare and unusual case of multiple hemangiomas of oral soft tissue and one intraossous hemangioma with phleboliths in mandible in a 45-year-old male.

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INTRODUCTION

Hemangioma is a term that encompasses a heterogeneous group of clinical benign vascular lesions that have similar histologic features. It is bening lesion, which is a proliferating mass of blood vessels and do not undergo malignant transformation. There is a higher incidence in females than male (Enzinger, 1995). According to histopathologic features of hemangiomas, capillary, cavernous, mixed types are available (Enzinger, 1995). Hemangiomas cannot be recognized at birth but arise subsequently during the first 8 weeks of life. They are the most common tumors of infancy, occurring in %5 to %10 of 1 year old children. The average age at diagnosis is 10 years, with 65% occurring in the first 2 decades of life (White, 2019). The majority of haemangioma involve the head and neck. However, they are rare in the oral cavity but may occur on tongue, lips, buccal mucosa, gingiva, palatal mucosa, salivary glands, alveolar ridge, and jaw bones (Enzinger, 1995; Gombos, 2008; Shafer, 2016).

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Clinically, haemangioma appears as soft mass, smooth or lobulated, and sessile or pedunculated and may vary in size from a few mms to several cms (Neville, 2016). They are usually deep red and may blanch on the application of pressure and if large in size, might interfere with mastication (Lone, 2017). Here, we report a rare case of multiple hemangiomas with phleboliths in oral cavity with a brief review on role of radiology in diagnosing such cases.

CASE REPORT

A male Patient aged 45 years reported to deptt. of dentistry with chief complaint of pain and swelling in left back tooth region since 1 month. History dates back to 1 month when Patient noticed a growth inside mouth on left cheek region which has gradually increased to present size. During chewing pain and bleeding occurs from the growth which subsides by itself. Patient gives history of multiple swellings in mouth since birth. On extraoral inspection, Diffuse swelling present on lower left 1/3rd of face measuring about 2.5 ×2.5 cm in its greatest dimensions (fig-1). Skin over swelling appears smooth with brownish pigmentation of skin and no visible pulsations, sinuses or fistulas. On Palpation extraorally, swelling is soft in consistency, non tender.

Fig. 1. Diffuse swelling on lower left 1/3rd of face

Fig- 2 swelling in gingivobuccal sulcus wrt 37,38

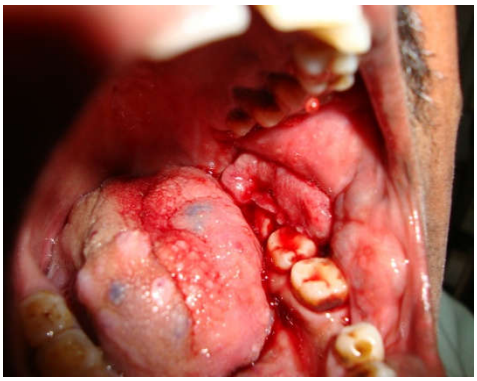

Fig-3 Bluish diffuse Swellings present on left and right buccal mucosa

Fig-4Bluish diffuse Swellings present on right lateral border of tongue and tip of tongue

Fig-5 Bluish diffuse Swellings present on ventral surface of tongue and floor of mouth

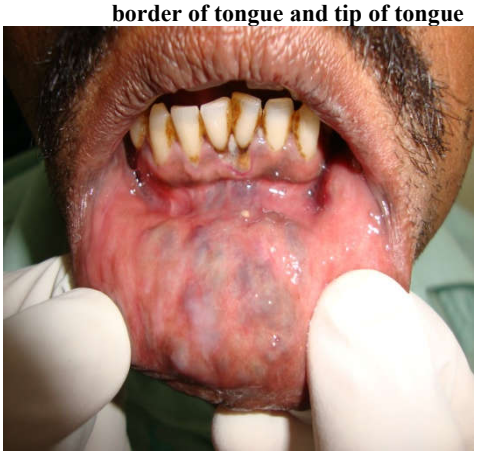

Fig-6 Bluish diffuse Swellings present on lower labial mucosa

Fig. 7. Pebbly irregular growths present one at posterior 1/3 rd and other purplish growth at anterior 1/3rd on dorsum of tongue

Fig. 8. IOPAR shows unilocular radiolucency distal to 33, 38 and enlarged inferior alveolar canal

Fig. 9. Mandibular occlusal radiographshowing multiple radiopacities(phleboliths) in floor of mouth

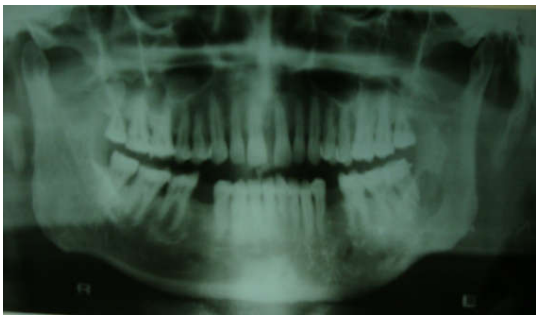

Fig. 10. OPG shows cystic radiolucency distal to 38 involving inferior alveolar canal and multiple oval radiopacities suggestive of phleboliths

Fig. 11. Ultrasonography of swelling revealed Hemangioma of left masseter extending in to tongue with multiple phleboliths

Intraorally there was Well circumscribed growth present on gingivobuccal sulcus wrt 37 ,38 , purplish red in colour measuring about 2.5×1.5 cm obliterating the buccal vestibule with irregular surface (fig-2). The Swelling was soft in consistency, tender with well defined borders and bleeding on probing was there from gingival sulcus. Bluish diffuse Swellings were present on left and right buccal mucosa (fig-3), right lateral border of tongue and tip of tongue (fig -4), ventral surface of tongue and floor of mouth (fig -5), lower labial mucosa (fig-6) were soft in consistency ,mobile , compressible , non pulsatile and non tender. Pebbly irregular growths were present on dorsum of tongue, one at posterior 1/3 rd and other at anterior 1/3rd of tongue measuring about 2×2cm (roughly) purplish in colour (fig-7). Tenderness was there on Percussion with respect to 37 and 38. Right and left submandibular lymph nodes were palpable, mobile, tender, single in no. oval in shape and soft in consistency. So provisional diagnosis of haemangioma with differential diagnosis of AV Malformation, Lymphangioma and Gingival carcinoma. Routine Blood Investigations – RBS, Hb%, CT,BT, TLC, DLC , ESR and INR were within normal limits.

In radiological investigations, IOPAR revealed mixture of normal and enlarged cancellous spaces with coarse and elongated trabeculae with respect to 36 ,37 along with unilocular radiolucency distal to 33, 38 and inferior alveolar canal was enlarged (fig-8). Mandibular occlusal radiograph was showing multiple radiopacities irregular in shape suggestive of phleboliths in floor of mouth (fig-9). OPG revealed coarse trabecular pattern in left body region of mandible alongwith a cystic radiolucency measuring about 2×1.5 cm , oval in shape distal to 38 involving inferior alveolar canal and multiple oval radiopacities suggestive of phleboliths present in soft tissues of face and neck (fig-10).

Ultrasonography of swelling revealed Hemangioma of left masseter extending in to tongue with multiple phleboliths. So final diagnosis was of multiple haemangiomas with phleboliths. For treatment and further management patient was referred to higher institution.

DISCUSSION

Hemangiomas are benign vascular lesions that cause symptoms such as mass, swelling, pain and discoloration (Jain, 2016). They occur on both skin and mucosal surfaces frequently in the head and neck region [5] but are rare in the oral cavity (Dhiman, 2015). Present case was unilaterally seen in head and neck area with multiple lesions in the oral cavity. Phleboliths are calcified thrombi found within vascular channels and occur frequently in the presence of hemangiomas or vascular malformations. They arise from injury to a vessel wall or result from stagnation of the flow of blood. Phleboliths generally cause no symptoms. Radiologically, they are seen either radiolucent or radiopaque. A fibrinous component is attached to the developing phleboliths and becomes calcified. Repetition of the process causes a layering effect and so phlebolith usually has a concentric ring or onion like appearance (Hemangioma Of Mandible, 2015). Mandible is a very infrequent location although when detected in the mandible, the greatest frequency of occurrence has been the body region, but condylar tumors have also been reported. In our case, similar location of occurrence was observed. It is usually asymptomatic although patient may present with signs and symptoms of slow-growing swelling of the bone, which may or may not cause facial asymmetry.[10] Other features include discomfort, oozing or pulsatile bleeding from the gingiva of teeth in the region of the lesion, mobile teeth, and accelerated exfoliation of teeth. In lesions with high vascular

pressure, patients often report a sensation of pulsation, and large lesions extending into adjacent soft tissues may have audible bruits (Jindal, 2010). Despite the benign nature of the lesion, paresthesia in the region is not uncommon. It is crucial to report that patients may not demonstrate any signs or symptoms. Failure to arrive at correct clinical diagnosis of an intraosseous hemangioma surgical intervention may lead to significant hemorrhage and even death (Aynalı et al., 2014). A definitive diagnosis from the clinical and radiographic features may not be possible without a biopsy. Removal of tissue for microscopic examination, however, carries with it the risk of uncontrollable hemorrhage and should be avoided. Angiography has proved to be a useful diagnostic tool when the features are prompting a diagnosis of hemangioma. This will be helpful in demonstrating the pressure of the vascular lesion as well as delineating the boundaries and arterial connections.[10] The important signs for the diagnosis of cavernous hemangiomas are bidigital palpation of the region and detection of disappearance of the blood on finger pressure and appearance of blood after removal of finger pressure. Furthermore, if the lesion has an arterial origin, pulse can be obtained by finger pressure. In ultrasound, color Doppler sonography has been developed to identify vasculatures and to enable evaluation of the blood flow, velocity, and vessel resistance together with surrounding morphology. It can be used for detecting the course of the facial artery and for detecting hemangiomas.[13] The treatment for benign vascular lesions include surgical excision with blade or laser, cryosurgery, injection of corticosteroids or sclerosant (sodium tetradecyl), radiotherapy, and embolization with steel coil, gel foam, silicone beads, or cyanoacrylate. Whether they should be followed up or treated depends on the patient's age and site and size of the lesion. These treatment modalities mentioned above have notable disadvantages including excessive bleeding, fibrosis, scarring, cosmetic and functional deficiency due to the long-term effect of steroid and radiation. However, surgical excision and cryosurgery are effective treatments for small lesions and superficial ones. The technique of circumferential (intratumoral) ligation takes advantage of the easy accessibility of the feeder vessels (An Isolated Phlebolith on the Lip, 2015; Gianfranco, 2014; Senthil, 2010).

Conclusion

As dental practioners, it is our duty to be consistent in our terminology and classification of vascular lesions in all our scientific writings and presentations in order to communicate effectively, understand its pathophysiology, promote research and develop newer therapeutics. When complications are likely or the threshold for intervention is uncertain, referral to an experienced specialist or a multidisciplinary vascular anomalies center may be advantageous.

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