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RESEARCH ARTICLE

INTRAORAL VENOUS MALFORMATION WITH PHLEBOLITHS – A CASE REPORT WITH REVIEW OF THE LITERATURE

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Abstract

Venous malformations formdue to disturbances during the developmental period of an embryo. Occasionally, associated with calcified bodies known as phleboliths. Pathological calcification of soft tissues is modulated by the deposition of calcium and other mineral salts. Phlebolith formation, is reported as a predictable feature of hemangiomas was first described in the splenic vein by Canstatt in the year 1843.The overall incidence of vascular anomalies is approximately 1 in 10,000 while children are more affected. They can occur anywhere in the body involving dermis, subcutaneous tissue, intraoral mucosa and skeletal tissue with an incidence around 40% in head and neck regions. They are arbitrarily distributed in various number and size, radiographs aid in detection of calcifications. Vascular malformations of oral cavity are not uncommon, with preponderance towards tongue, lips, buccal mucosa, gingiva, palatal mucosa and oropharynx.Treatment of vascular malformations are managed by more than one modalities like surgical excision, laser therapy and sclerotherapy. Herein we are reporting a rare case of an intraoral venous malformation with phleboliths.

Introduction:-

Venous malformations form due to disturbances during the developmental period of an embryo.Occasionally they are associated with calcified bodies known as phleboliths. Injury to a vessel wall or torpidity in the blood flow triggers thrombus formation.Pathological calcification of soft tissues is modulated by the deposition of calcium and other mineral salts.[1]Phlebolith formation, is reported as a predictable feature of hemangiomas was first described in the splenic vein by Canstatt in the year 1843. The overall incidence of vascular anomalies is approximately 1 in 10,000 while children are more affected. They can occur anywhere in the body with an incidence around 40% in head and neck regions. They are arbitrarily distributed in various number and size,, radiographs aid in detection of calcifications. [2,3,4]

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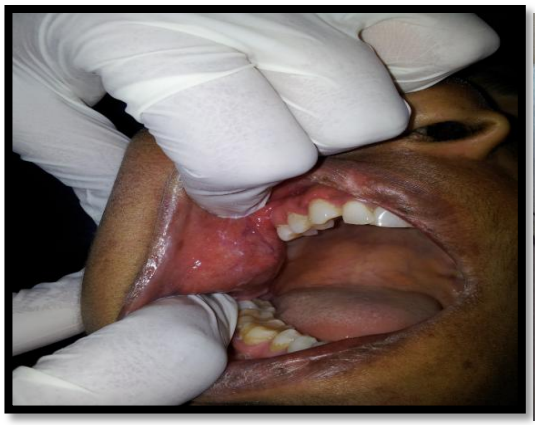
Vascular malformations of oral cavity are not uncommon,with preponderance towards tongue, lips, buccal mucosa, gingiva, palatal mucosa and oropharynx. On literature search few cases in buccal mucosa, masseter,temporalis, mentalis muscle and floor of the mouth were found to be present as swellings.[2,3]Treatment of vascular malformations are managed by more than one modalities like surgical excision, laser therapy and sclerotherapy.[5,6] The present case of venous malformation with phleboliths was found intraorally on the buccal mucosa of a 45-year-old woman.

Clinical Details:-

A 45 years old woman presented with swelling and painin the right cheek of8months' duration. No other relevant dental or medical history were found. History of aninjury to the right cheek3 years back withincrease in the firmness of swelling over the years. Clinical examination revealed bluish tinge on the right buccal mucosa extending from the vestibule close to first molar to the anterior border of the ramus.Submucosal mass was firm in consistency, compressible and with no pulsation. Submandibular lymph nodes were palpable, parotid and submandibular salivary glands were normal. It was provisionally diagnosed as sialolithiasis.

Gross details &Histopathology: -

Excisedspecimenwasyellowish blue in colourwith multiple whitish hard nodules. Histopathology revealed loose adipose tissues with interconnected numerous dilated blood vessels.Thrombus formation with dystrophic calcifications were present inside these dilated veins. Thickened veins displayed collagen deposition on the wall lined by flat endothelial cells along with neutrophils, lymphocytes and plasma cells. The diagnosis indicated avenous malformation with phleboliths.



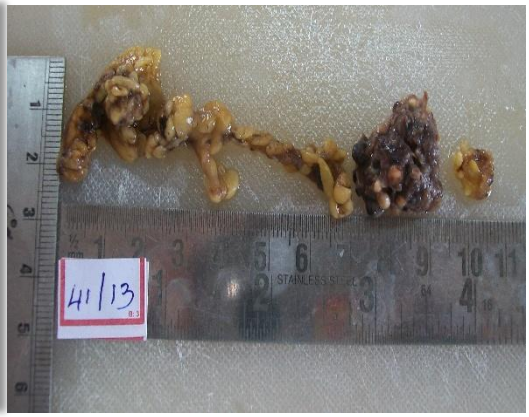
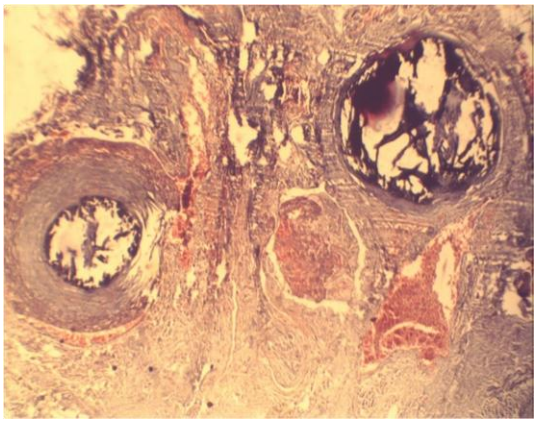


Fig 1:- Swelling in the right buccal mucosa.**Fig 2:-** The gross specimen yellowish blue in colour measuring around 10cmx2.5cmx2cm.



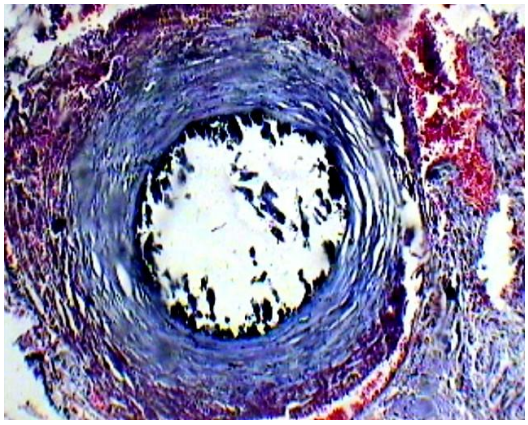


Fig 3:- Numerous interconnected blood channels with thrombus formations(4x Magnification, Hematoxylin& Eosin).

Fig 4:- A large dilated vein with phleboliths(10x Magnification, Masson's Trichrome).

Discussion: -

Venous malformations also known as venous hemangioma, cavernous hemangioma or cavernous angioma are rather old terms used quite often in the earlier literatures. These are present at birth but hardly make their presence obvious. The classification of hemangiomas and vascular malformations was clearly emphasized by mulliken et al.[7,8] It is based on their biological characteristics. Disturbances in vasculogenesis and angiogenesis leads to defects in the capillaries, veins, arterioles, lymphatics or combination of vessels.They are typically divided according to the blood flow into low-flow (capillary malformation, lymphatic malformation, and venous malformation) and high-flow lesions (arteriovenous malformations, arteriovenous fistulae). These have become the source of interest in the study of embryonic angiogenesis. Their presence is usually felt later in life. They accelerate during puberty, pregnancy as well as trauma like in our case. [9,10,11]The lesion was present in the buccal mucosa, for years till the trauma triggered it to expand with inflammation and pain.

Vascular anomalieshave a propensity to occur in the skin, mucosa, subcutaneous tissue and muscle. Phleboliths associated with vascular anomalies in the maxillofacial region were discovered by Kirmission in 1905. They are more characteristic of low-flow vascular malformations. According to the theory adopted by Ribbert, phlebolith formation begins with intravascular thrombus formation and is followed by progressive lamellar fibrosis.[12,13]In initial diagnosis, radiographs are the essential tools. Based on the anatomic considerations, differential diagnosis depends on the contents in and around lesion, buccal pad of fat, minor salivary glands, lymph nodes, Stenson's duct, accessory parotid gland, numerous vessels and nerves. Since the mass was localized to an area for many years and not invading the other structures, malignancy was ruled out. Though major salivary gland functions were normal possible shortlisted differential diagnosis includedsialolithiasis as bluish tinge of the mucosa was visible. Other possibilities were lipoma, vascular malformations, lymph node infections, neurofibroma,reactive soft tissue proliferation due to trauma, bacterial or fungal infections. Intraoral lipomas are seen most common on the buccal mucosa,with predilection for adult males. They are found to be soft and nodular masses sessile or pedunculated.[14]Venous malformations are present from birth but patient may be without symptoms unless or otherwise due to trauma or infection. Found intraorally more on the buccal mucosa, masseter or tongue. Colour of the lesion are clinically observed varies depending on the depth, from colourless to bluish tinge visible on the mucosa. Compressible lesions help in identifyingvenous malformations from lymphatic malformations. Calcificationsrequires differentiation mainly from sialolithiasis, phleboliths, calcified lymph nodes, healed acne lesions.[15,16]Histopathologyrevealedphlebothrombotic areas in numerous large dilated vessels with collagen formation, flat endothelial cells and haphazard channels.It was verified by Masson's trichrome special staining. Location, extent, growth rate, accessibility, patient's age, and aesthetics plays an important role in treatment options. Interstitial laser therapy has found to be effective for capillary and venous malformations. Embolization solely or accompanied with surgery holds good for high flow vascular malformations.[17,18,19]

Conclusion:-

Phleboliths in vascular malformations are calcified bodies, not many intraoral cases have been found in the literatures.Though clinical diagnostic hypotheses were numerous to deal in the present case,histopathologyconfirmed the diagnosis. Since, now the old terminologiesare not in vogue, venous malformations are identified well, as clear entities on their own.

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