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Case Report

Cavernous hemangioma of buccal mucosa: an unusual presentation: case report and review of literature

Mahesh S. G., Rukma Bhandary, Deepalakshmi Tanthry, Vishwas K. Pai, Sapna M.*

Department of Otorhinolaryngology, AJIMS, Mangalore, Karnataka, India

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***Correspondence:**
Dr. Sapna M.,
E-mail: sapna.mallikarjun@gmail.com

ABSTRACT

Cavernous hemangiomas are benign vascular malformations which arises during first 8 weeks of life. Most of the Cavernous hemangiomas, about 90% undergo spontaneous involution, with a small proportion of cases involving intervention. Cavernous hemangiomas can affect 1 in 200 people hence the incidence of cavernous hemangioma of buccal mucosa accounting for much rare of an entity. Here we present one such rare and interesting case which required a surgical intervention as a modality of treatment. 45-year-old male patient presenting to us with complaints of swelling over left cheek for 6 months. After thorough clinical and radiological evaluation, the mass was surgically resected and specimen was sent to HPE- Reported as cavernous haemangioma. The incidence of cavernous haemangioma of being 0.5%. The aim of this case report was to discuss the journey of one such rare incidence of cavernous hemangioma of buccal mucosa with surgical intervention being the modality of treatment.

Keywords: Vascular tumors, Cavernous hemangiomas, Resection, Buccal mucosa

INTRODUCTION

Hemangiomas are thought to be congenital vascular malformations arising from abnormally differentiated and proliferating endothelial cell network.[1] The tumor is present in the first decade of life in about half of all cases, with the vast majority being detected before the end of the third decade of life.[1] There is no sex predilection.[1] Although hemangiomas can occur in the body, 15% are in the head and neck region and only 1% arise in skeletal muscle.[1]However, of those that do arise in skeletal muscle, the masseter muscle represents the most common site followed by trapezius, sternocleidomastoid, temporalis, mylohyoid, mentalis, buccinators, lip, tongue, and salivary glands.[1]

Cavernous hemangiomas are less likely to spontaneously involute.[2] These benign, vascular lesions are slow growing and can manifest as a painless, progressively mass.[2] The scalp, face and neck are the most common sites, but these tumors have been found in the liver and other organs.[2] Cavernous malformations are vascular malformations with low flow hence they are are 'occult' to angiography.[2] On CT scan, one may see the evidence of hemorrhage of various ages and with contrast administration the lesion itself may enhance.[2] MRI offers the most sensitive means of suspecting a diagnosis of cavernous malformation.[2] In the world of enigmatic arteriovenous malformations we are here to discuss about cavernous hemangioma of head and neck region emphasising on the buccal mucosa as the foci of discussion.

CASE REPORT

45-year-old male patient presenting to ENT OPD with complaints of swelling over left cheek for 6 months. The swelling was insidious in onset, slowly progressive. The patient did not have any history of difficulty in chewing

and swallowing, history of numbness of face and swelling in other part of body. On examination, 3×4 cm size single swelling was involving left cheek. It was extending from zygomatic process of maxilla superiorly, angle of mandible inferiorly, anteriorly from angle of mouth, posteriorly 5 cm from tragus. The overlying skin was normal and on palpation local temperature was raised. The skin was pinchable, margins were not demarcated, firm in consistency with small size multiple hard area present and compressible. On auscultation, bruit was audible. The ultra-sonogram of the mass was done and was reported as? sarcoma or malignancy. The patient underwent excision biopsy of mass under general anaesthesia with suspected diagnosis of vascular malformation. The control on feeding vessel was taken and mass removed in toto and sample was sent to Histopathological examination which was reported as cavernous haemangioma. The patient was disease free even after 6 months of follow-up.

Post-operative follow up

Patient was followed up post operatively for 6 months with minimal scar noted without any recurrence.

Patient perspective

It was satisfactory.

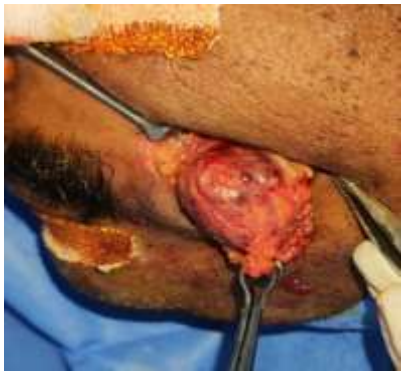

Figure 1: Intra-operative figure showing the mass with vascular malformation.

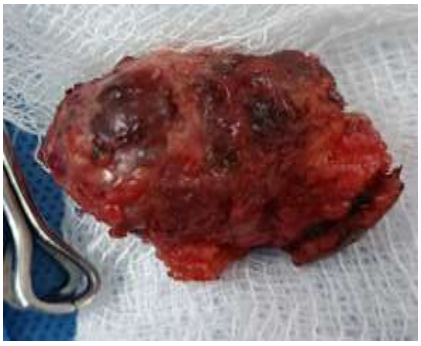

Figure 2: Gross specimen removed in-Toto consisting of dark brown mass measuring 3.2 cm in diameter. Cut section shows homogenous dark brown and blackish areas.

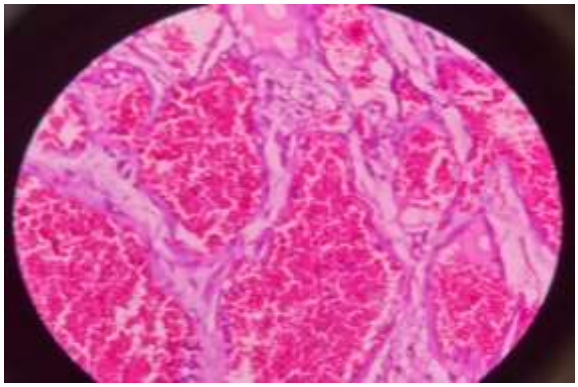

Figure 3: Tumor tissue made up of multiple irregular large dilated vascular spaces lined by single layer of endothelial cells.

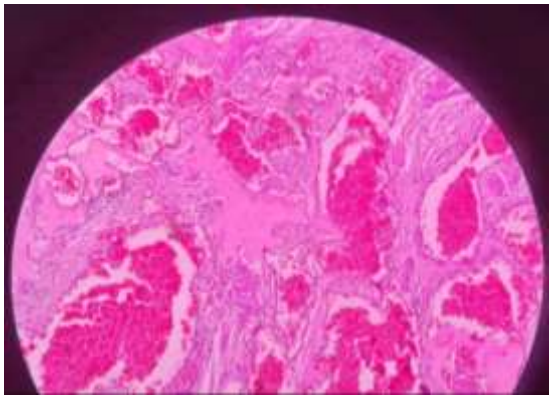

Figure 4: These are surrounded by thin fibro-collagenous stroma showing small foci of haemorrhage.

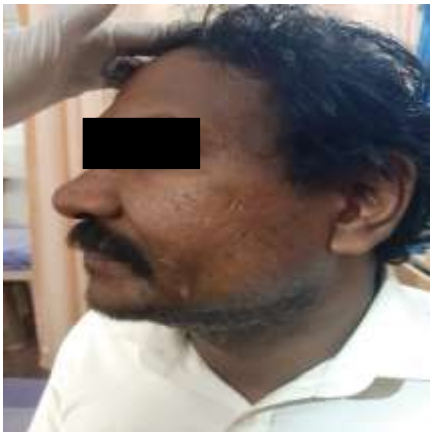

Figure 5: Post-operatively minimal scar was noted.

DISCUSSION

William Hunter in the mid-eighteenth century first described vascular anomalies.[3] Hemangiomas are congenital, benign, well-circumscribed malformations of the vasculature.[2] Vascular anomalies are lesions arising from the arterial and/ or venous and/or lymphatic circulation.[3]

Arterial malformation are dilated, overlapping and tortuous arteries having a coil-like appearance and/or a collection of arterial loops without any venous component.[3] They are also described as 'high flow lesions'.[3] Venous malformation are commonest type of vascular malformations caused by ectatic venous channels, also called 'low flow lesions'.[3]

Capillary malformations are commonly known as, 'port wine stain'.[3] It is a flat, well-defined vascular stain of skin seen early in development when vessels of skin form abnormally which can increase in size and give a nodular appearance as a late presentation.[3] Lymphatic malformation are lesions containing fluid-filled spaces or channels, thought to be caused by abnormal development of the lymphatic system.[3]

It is commonly known that skin overlying a hemangioma shows increased vascularity, giving the lesion reddish-blue discoloration or even hyperthermic change.[4] Hemangiomas of the head and neck are usually focused on cutaneous surfaces, but can be located on mucosal surfaces as well.[5] Involvement of the skull base, especially of the infratemporal fossa, is rare, but usually occurs by extensions of intramuscular or osseous lesions where intra osseous lesions have been reported less than 1%.[5]

Haemangiomas have currently 2 dominant theories, although unclear.[3] These are: (1) endothelial cells are formed from deranged placental tissue present in foetal soft tissues during gestation; and (2) stem cells and endothelial progenitor cells which give rise to haemangiomas, are found in circulation of patients with haemangiomas.[3]

The pathogenesis of arterio-venous malformations (AVMs) is also not clearly understood.[3] A defect in vascular stabilisation is known to potentially cause AVMs.[3] Abnormal levels of matrix metalloproteinases and proangiogenic factors are involved in the pathogenesis of haemangioma.[3] Genetic errors involving growth factor receptors are also known to influence the development of these lesions.[3]

Haemangioma in the maxillo-facial region is uncommon with only few cases reported in the medical literature.[6] As far as the incidence is concerned, intra-osseous.[6] Hemangiomas constitutes 1% of the all Osseous tumors.[6] Superficial cavernous hemangiomas are friable and easily infected if the skin is broken.[2] They vary in size from punctate to several centimeters.[2]

Histologically, vascular anomalies are seen as a localised increase in vasculature with abnormal tortuosity and eenlargement.[3]

Superficial haemangiomas can appear as raspberry-coloured birthmarks or reddish discolouration of the skin.[3] The bright red strawberry-like classic appearance may not be seen in deeper lesions involving sub-cutaneous tissues.[3] Compound lesions involving superficial and deeper tissues are also seen.[3] The clinical appearance may vary depending on the depth of tumour.[3] Growing haemangiomas can show organ involvement and cause ulcerations, bleeding, hearing problems, vision changes, difficulty in mastication, dysphagia and dyspnea.[3]

Other pathologic lesions are usually confused in the differential diagnosis, like neoplasms in parotid gland, benign muscular hypertrophy especially related to the masseter, or congenital cystic.[4]

Radiological assessment

Ultrasonogram is the least invasive modality of imaging available for assessment of vascular anomalies.[3] It is used as a baseline investigation for superficial head and neck vascular lesions.[3] In addition to the above, the ultrasonography has a high sensitivity in diagnosing these lesions.[3] The disadvantages of the ultrasonography are its limitation in detecting deeper soft tissue and bony lesions.[3] Doppler ultrasound is used to demonstrate high-flow lesions.[3] Flow towards the ultrasound transducer is seen in red and away from the transducer seen in blue.[3] The arterial feeder is usually identified by increased colour flow, high doppler shift and low resistance.[3]

CT is used to diagnose bony lesions, provide cross-sectional details and detect the presence of calcifications like phleboliths in venous malformations and haemangiomas.[3]

MRI is the most preferred and accepted imaging for diagnosing and monitoring soft-tissue vascular lesions of head and neck.[3] Its superior contrast resolution, in- depth soft-tissue assessment and non-exposure to ionising radiation, makes it the investigation of choice for these lesions.[3]

Management

The treatment options include surgery, irradiation, laser therapy, cryotherapy and instillation of sclerosing agents.[2] The most effective treatment is a wide surgical excision.[2] Surgery is indicated for large tumors that invade adjacent structures and cause functional derangement, disfigurement or pain.[2] In haemangiomas, a 'wait and observe' strategy is preferred. Involution is seen in more than 85% of patients.[2] Medical line of management includes propranolol and triamcinolone.[3]

Surgery is considered between 2 and 4 years of age after having attempted medical treatment to minimise deformity involving eyes, nose and lips and other areas of face.[3] Smaller-sized lesions are almost always excised completely. In contrast, larger-sized lesions are mostly debulked and need multiple procedures.[3]

According to the author's classification, vascular malformations were categorised into five types based on

their anatomy and depth of location in the head and neck.[3] This is a good guide for selecting the type of surgical management and reconstruction (a) in type-I, superficial lesions require excision of skin or mucosa.[3] Local or regional flaps have been used in reconstruction of the residual defect; (b) type-II, sub-mucosal lesions require complete excision after elevation of skin flaps; (c) type-III, lymphovenous malformations or venous malformations involve salivary glands and are excised along with the affected gland; (d) type-IV, intra-osseous lesions require excision with removal of involved bone and reconstruction of the residual defect as necessary; and (e) type-V lesions involve deep visceral spaces, such as the parapharyngeal or infra-temporal fossa and usually require mandibular access osteotomy for complete exposure and complete removal of the lesion.[3]

CONCLUSION

To narrow down such huge array of topic into a small brief closure is quite exigent. In the world of vascular anomalies, to identify the lesions, categorizing them and early detection and plan of management can avoid the reccurence. With surgical excision being the modality of treatment in the small or large long standing hemangiomas, with evolving array of sclerosing agents, it is important to keep the side effects in the lane, to upgrade the prognosis and line of cosmetic track of treatment with better outcomes and results and least or even nil rate of treatments.

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