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Case Report

Hemangioma of the buccal mucosa treated with sclerotherapy: A rare case report

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ABSTRACT

Hemangiomas are common benign vascular anomalies that can occur in various locations throughout the body, including the oral cavity. The lesions are generally asymptomatic and exhibit a variation in the size and colour. Here, we present a case report of a hemangioma located in the buccal mucosa of a 40-year-old female patient who presented with asymptomatic reddish purplish swellings in the buccal mucosa, which had gradually increased in size over the last 6 months. Clinical examination and imaging studies confirmed the diagnosis of a hemangioma. 1 ml of 3% of sclerosing agent (sterol; sodium tetradecyl sulphate) mixed with lignocaine HCL was injected in the periphery of the lesion, once a week for 3 weeks, and periodic follow-up evaluation revealed a marked reduction in the lesion size. This case demonstrates the feasibility and efficacy of sclerotherapy as a minimally invasive treatment option for hemangiomas of the buccal mucosa, particularly in cases where surgical excision may be technically challenging or associated with significant morbidity.

1. Introduction

Hemangiomas are common benign, non-reactive vascular tumours, characterized by the presence of numerous blood vessels. While around 60-70% of hemangiomas are typically found in the head and neck area, their occurrence within the oral cavity is exceptionally rare.[1]

The term "hemangioma" is derived from the Greek words "Hema" meaning blood, "angeio" meaning vessel, and "oma" indicating tumour. Clinically, hemangiomas may present in two forms, i.e., Infantile (IH) or congenital hemangiomas.[2,3] Infantile hemangiomas (IHs) typically show positivity for glucose transporter protein-1 (GLUT1), distinguishing them from congenital hemangiomas. Apart from this genetic difference, several distinct features aid in distinguishing between the two types.[4] IHs emerge within the initial two months of life, rapidly expanding between six months and one year, followed by a gradual regression.[5] In contrast, congenital hemangiomas are evident at birth and may become symptomatic due to growth, infection, hormonal changes, or traumatic injury.[4]

Hemangiomas typically manifest as a benign, soft, and often compressible mass, displaying variations in size and coloration that can range from red to bluish-purple. While they are generally asymptomatic, larger lesions may cause discomfort, impair oral function, or lead to spontaneous bleeding.[2]

While the precise etiology remains uncertain, it is thought that hemangiomas arise from a dysregulated vascular development during embryonic stages.

The diagnosis primarily relies on patient's medical history and clinical examination, supplemented by imaging techniques such as ultrasonography (USG) or magnetic resonance imaging (MRI).[6]

There are no established treatment protocols for hemangiomas, and recommendations may include medical,

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interventional, or surgical approaches. Treatment is typically carried out for patients experiencing either aesthetic or functional concerns.[7] The choice of treatment depends on factors such as the size, location, and symptoms of the lesion. Various treatment strategies include observing for spontaneous regression, surgical excision, radiation, laser therapy, sclerotherapy, cryosurgery, and feeder vessel embolization.[8]

Although typically benign, hemangiomas of the buccal mucosa may require treatment to relieve symptoms or for cosmetic purposes. This article aims to present a case report of hemangioma in the buccal mucosa treated with sclerotherapy.

2. Case Presentation

A 40-year-old female patient presented at our Outpatient Department with asymptomatic reddish-purplish swellings in the buccal mucosa. The patient noticed the swellings a year back, which gradually increased in size over the past 6 months. Her medical and family history was non-significant. There was no ulceration or bleeding associate with the lesion.

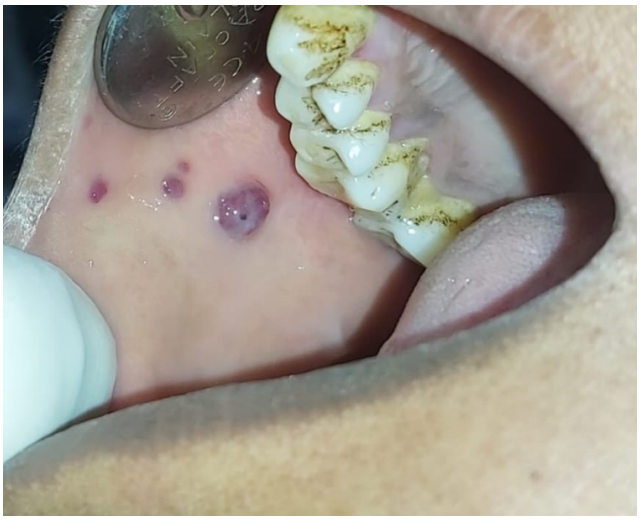

Figure 1: Dome shaped reddish-purplish lesions on thebuccal mucosa

Physical examination revealed no noticeable facial asymmetry, and no abnormalities were observed in the cervical lymph node and neck regions. Clinical examination of the right buccal mucosa revealed the presence of three well defined, sessile, dark red to purplish dome shaped swellings with an intact mucosa extending along the occlusal plane, roughly measuring 1.5cm X 2cm in its greatest dimension (Figure 1). On palpation, the swellings were non-tender, non-pulsatile, soft in consistency, and exhibited a positive diascopy test. (Figure 2). A thorough general examination ruled out the presence of any similar

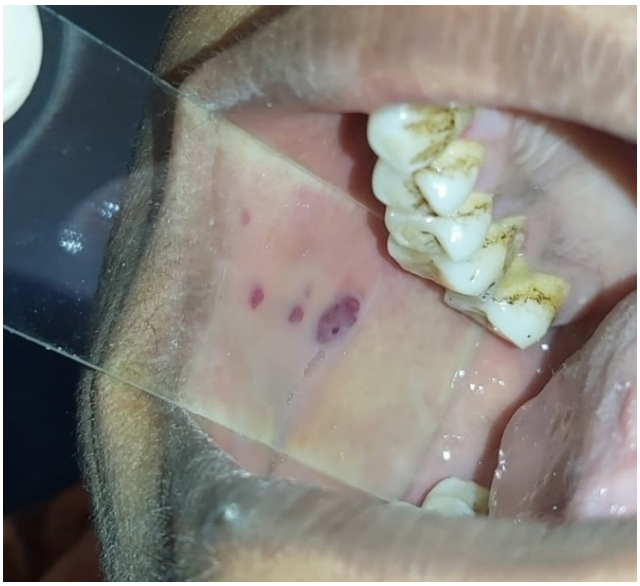

Figure 2: Blanching of the lesion on applying pressure (PositiveDiascopy Test)

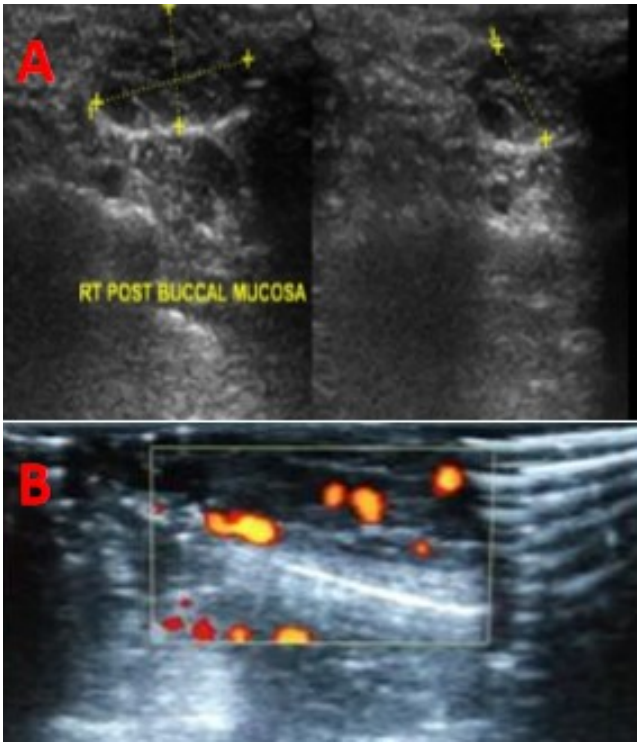

Figure 3: A & B: Ultrasonography revealing hypoechoic, soft tissue masses with a venous type of vascularity.

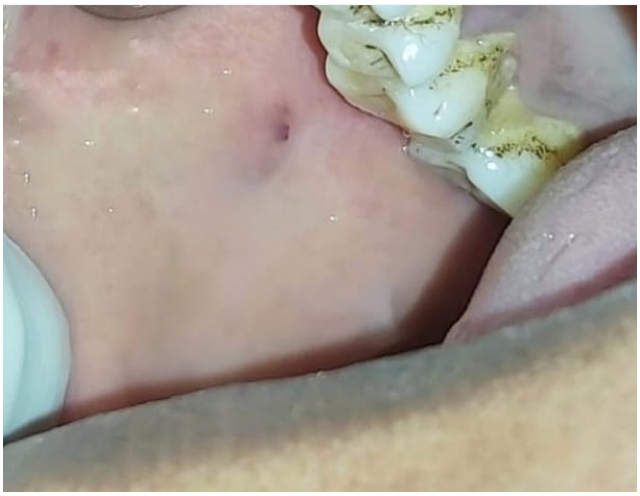

Figure 4: Resolution of the lesions after injections ofsclerosing agents

Table 1: Differentiating features between hemangioma and vascular malformations.

Hemangiomas	Vascular malformation
A hemangioma may or may not be present at birth	Vascular malformation is always present at birth
They are true benign neoplasm of endothelial cells	Are localized defects of vascular architecture that results in formation of abnormal torturous and enlarged vascular channel
Females are more commonly affected 3:1	Vascular malformations show no gender predilection
Hemangiomas are also known as port-wine stain, strawberry hemangioma, and salmon patch	Vascular malformations are also known as lymphangiomas, Arteriovenous malformation, and vascular gigantism
Grows faster often faster than the child's growth	Enlarges proportionately with growth of the child
Over time they become smaller (involute) and lighter in colour	They do not involute spontaneously and may become more apparent as child grows
Mast cells known to play role in neo-angiogenesis, increases during proliferating phase	No increase in mast cells

swelling in the body.

Considering the patient's medical history and clinical examination, hemangioma or vascular malformation were listed as the possible differential diagnosis. The patient was subjected to ultrasonography imaging and hematological investigations. All the hematological investigations were in the normal range. A hypoechoic, soft tissue lesion measuring 1.4 x 0.7 x 0.5 cm in the right buccal mucosa with a venous type of vascularity and no evidence of calcifications was observed on color-doppler ultrasonography. The mass displayed a homogeneous echotexture, indicating a benign nature, with no evidence of invasion to any adjacent structure.(Figure 3) The ultrasonography imaging was suggestive of hemangioma of the buccal mucosa.

Considering the patient's comfort and minimizing the risk of bleeding, sclerotherapy was regarded as the preferred treatment. 1ml of 3% sclerosing agent (Sterol; sodium tetradecyl sulphate) mixed with lignocaine HCL was injected in the periphery of the lesion, once a week for 3 weeks with an insulin syringe. The lesion exhibited a marked reduction in the size with no associated adverse effects (Figure 4). The patient was periodically reviewed once in 3 months, and no recurrence was observed during the one-year follow-up.

3. Discussion

The diverse clinical manifestations of vascular deformities have led to uncertainty in their diagnosis and classification. Mulliken and Glowacki (1982) aimed to address this ambiguity and were the pioneers in establishing a systematic terminology for vascular deformities.[9] Based on the pathological characteristics (endothelial cell turnover), these vascular deformities fall into two categories: (a) Vasoproliferative neoplasm (haemangiomas) and (b) vascular malformations (VMs). VMs are structural anomalies affecting the veins, lymphatics, capillaries, and arterioles, expand proportionately with the child's growth, and exhibit lower endothelial cell turnover (less proliferation and mitosis) compared to vasoproliferative neoplasms. Growth of vascular lesions occurs due to changes in blood flow and pressure, dilation of vascular channels, and proliferation of collateral vessels.[7]

While haemangiomas and VMs may exhibit clinical similarities, they possess unique histopathologic and histochemical features that aid in establishing a definitive diagnosis.[3](Table 1)

Differences betweenhemangioma and vascular malformations.

Oral hemangiomas (OHs) are benign vascular tumours resulting from endothelial cell proliferation and typically arise within or around the oral cavity.

While the head and neck region account for 60 to 70% of hemangiomas, OHs are relatively infrequent and typically seen on the lips, tongue, buccal mucosa, and palate.[5,9]

The precise etiology remains uncertain, and the pathogenesis of OHs is influenced by a complex interaction of several factors including genetic predisposition, hormonal changes, dysregulated angiogenesis during embryonic development, placental factors, and inflammatory pathways.[10,11]

OHs exhibit considerable variation in the clinical appearance and may appear as bright red to purplish superficial nodules to deep submucosal mass, pinpoint to several centimeters in diameter. The head and neck region

is the primary location for most superficial soft-tissue hemangiomas, whereas deep-seated hemangiomas are more frequently found in the trunk and lower extremities.[12]

The patient may be either asymptomatic or may experience discomfort, facial asymmetry, bluish discoloration, bleeding, pulsations, tooth mobility, mobile teeth, premature tooth shedding, and tooth agenesis. Large lesions may cause a pulsating sensation, audible bruits upon extension into the soft tissue, and blanching under pressure. Patients may occasionally experience paresthesia in the affected area. In some cases, hemangiomas may exist without any outward signs or symptoms.[13] A salient feature of oral hemangiomas is blood seeping from the sulcus. Moreover, the affected teeth may exhibit a pumping motion when pressure is applied and released (pumping tooth sign).[7]

Diascopy, which involves applying pressure with a finger or glass slide to an oral lesion to observe branching, is a physical examination technique useful for distinguishing vascular lesions (which blanch) from purpura (which do not blanch). Additionally, oral hemangiomas may become more noticeable and darken when the head is lowered or when pressure is applied to the abdomen.[14,15] Although superficial hemangiomas can be readily detected through clinical examination, establishing a definitive diagnosis for deep hemangiomas may be more challenging. In such diagnostic scenarios, advanced imaging techniques such as Doppler ultrasound (USG) or magnetic resonance imaging (MRI) are essential for aiding diagnosis. Moreover, if radiographic features are deemed insufficient, histopathological evaluation is regarded as the most precise and dependable diagnostic approach for oral hemangiomas.[1]

Hemangiomas frequently exhibit spontaneous regression, and treatment is generally reserved for complex cases, such as ulceration, profuse bleeding, functional impairment, and facial disfigurement.[16] Various treatment modalities have been suggested for managing hemangiomas, including non-selective beta-blockers (such as oral propranolol and topical timolol), intralesional corticosteroid injections, laser photocoagulation, cryotherapy, excision, radiation, sclerotherapy, embolization, and chemotherapy.[7,16]

Hemangiomas of the buccal mucosa may pose therapeutic challenges due to their intraoral location, and the potential impact they may have on aesthetics, function, and patient comfort. Surgical excision has been the primary treatment approach for OHs, however, it can result in significant morbidity, including bleeding, nerve injury, and scarring, particularly in cases involving larger or deeply infiltrating hemangiomas.[17]

Sclerotherapy has evolved as a promising intervention for vascular abnormalities, including hemangiomas, It induces thrombosis within the vascular channels, resulting in subsequent fibrosis of the lesion. This minimally invasive method provides several benefits, such as avoiding surgical trauma, reduced risk of bleeding or infection, and preserving surrounding healthy tissues.[8] Sodium morrhuate, sodium tetradecyl sulphate (STS), sodium psylliate, ethanolamine oleate, and polidocanol (aethoxysclerol 3%, 1% or 0.5%) are the commonly employed sclerosing agents. Polidocanol (aethoxysclerol) and sodium tetradecyl sulphate (STS) are commonly used agents that causes regression of the lesion by triggering a localized inflammatory reaction. This reaction leads to obliterative thrombosis of the hemangiomatous space and subsequent fibrosis of the endothelial spaces.[17]

In our case, the patient presented with a well-defined hemangioma in the buccal mucosa. Sclerotherapy was chosen as an alternative approach, aiming to achieve lesion regression while minimizing morbidity. The procedure was performed under local anaesthesia, ensuring patient comfort and facilitating outpatient management. Follow-up assessments revealed a significant reduction in the size of the hemangioma and improvement in symptoms, consistent with successful sclerotherapy outcomes reported in the literature.

Although sclerotherapy presents numerous benefits, such as its minimally invasive approach and favourable cosmetic results, it does have its limitations. The efficacy of sclerosing agents may vary based on the type, size, and location of the lesion, and multiple treatment sessions may be necessary to attain desired outcomes. Moreover, it is essential to carefully weigh the risk of potential complications, such as tissue necrosis, scarring, or nerve damage.[18]

The prognosis for hemangioma is favourable as it typically does not recur or undergo malignant transformation following appropriate treatment.[19]

4. Conclusion

Hemangiomas are characterized by rapid endothelial cell proliferation during early infancy, followed by gradual involution. They are frequently seen in the head and neck region, although, oral hemangiomas are rare. Sclerotherapy presents a promising treatment option for managing haemangiomas in the buccal mucosa, providing a minimally invasive alternative to surgical excision. However, careful patient selection, individualized treatment planning, and close monitoring are essential to ensure optimal outcomes and minimize the risk of complications.

5. Source of Funding

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6. Conflict of Interest

None.

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