CASE REPORT

Sinusoidal Haemangioma of the Tongue: Case Report and Review of Literature

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Abstract:

Oral haemangiomas are benign vascular tumors that can be classified as cavernous haemangioma and capillary haemangioma. Sinusoidal Haemangioma is a rare variant of cavernous haemangioma and is extremely infrequent in oral region. It represents only 1% of all haemangiomas and occurrence in the intramuscular location is unusual. Here we report the case of 5 year old female patient with swelling on the left dorsal surface of the tongue which was diagnosed histo-pathologically as sinusoidal haemangioma. Even though sinusoidal haemangioma has distinctive histopathological characteristics, many at a time these lesions are misdiagnosed as cavernous haemangioma.

Key words: Sinusoidal Haemangioma, bluish red swelling, cavernous haemangioma, sinusoidal pattern

Introduction

Haemangioma was first described by Liston in 1843.[1] They are a group of hemartomatous disorders with either reactive or neoplastic origin. Haemangiomas accounts for nearly 7% of all benign tumours. The haemangioma usually appear 2-4 weeks after birth, develops quickly until the age of 6-8 months, and then progresses slowly. By the age of 5-8years they start to involute and spontaneously regress in almost 70% cases.

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Haemangiomas may arise as superficial or deep lesions.[2] The superficial lesions are easy to diagnose but there is little chance of diagnosing deep lesions as they are asymptomatic.

As per the classification published by Mulliken and Glowacki in 1982[3], the International Society for the Study of Vascular Anomalies (ISSVA) has provided guidelines to differentiate vasoformative tumours into haemangiomas and vascular malformations. Haemangiomas are classified histologically as capillary and cavernous forms.

In recent years there is a huge advancement in identifying and categorizing many rare vascular tumors. One of which is sinusoidal haemangioma, a rare variant of cavernous hemangioma.[4] The clinical appearance is similar to conventional haemangiomas but histologically it may mimic well differentiated angiosarcoma.

Here we report a rare case of sinusoidal haemangioma of tongue in a five year old female. The features of Sinusoidal haemangioma can lead to diagnostic pitfalls,

thus we consider it is useful to report our case along with review of literature.

Case Report:

A 5 year old female patient reported to the department with the chief complaint of a swelling on the dorsum of the tongue. Intraoral examination revealed an ill-defined solitary swelling with bluish red colour, approximating 2 x 2 cm on the left posterior aspect of tongue. The swelling was non tender and reducible but slowly fills back. Excisional biopsy was performed.

On gross examination of the surgical specimen, a greyish-white mass with irregular surface, measuring about 1.5cm X 1cm X 1cm was noted. Microscopic examination of the hematoxylin and eosin stained section revealed stratified squamous surface epithelium with spongiosis. Connective tissue stroma exhibits numerous large interconnecting vascular spaces. Dilated vascular spaces are lined by a thin layer of flattened endothelium with areas of pseudo-papillary pattern demonstrating a characteristic sinusoidal appearance. Multiple dilated vascular channels filled with organizing thrombi was noted and it also exhibits focal calcification with in the channel. There was no significant inflammation or nuclear atypia distinguished. With these histopathological features we came to a diagnosis of Sinusoidal haemangioma of tongue.

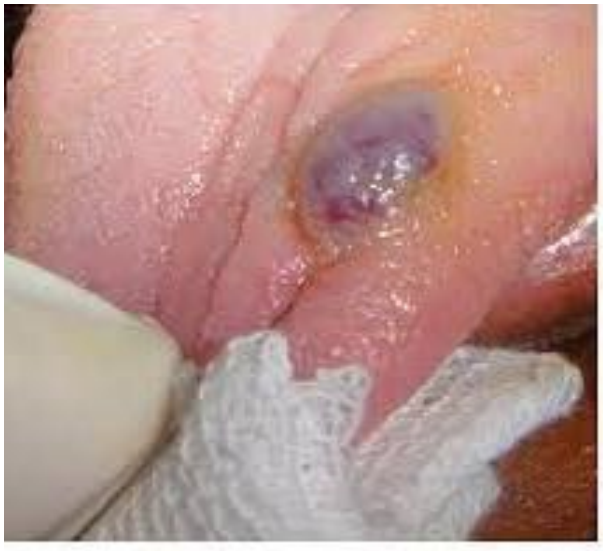

Fig 1: Bluish red raised lesion on the left lateral surface of tongue

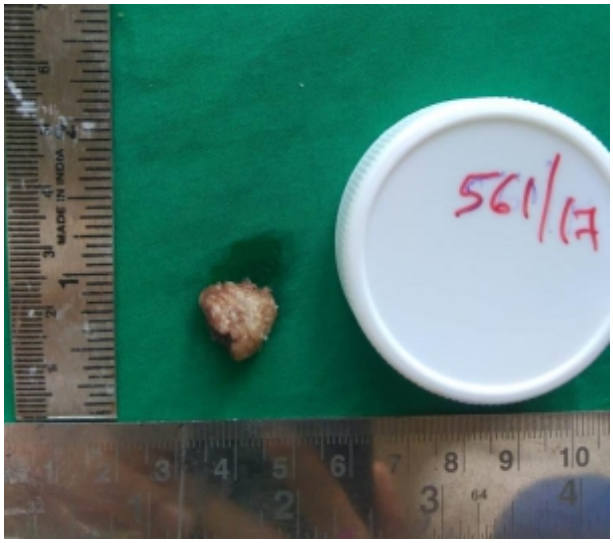

Fig 2: Irregular surface of the gross specimen

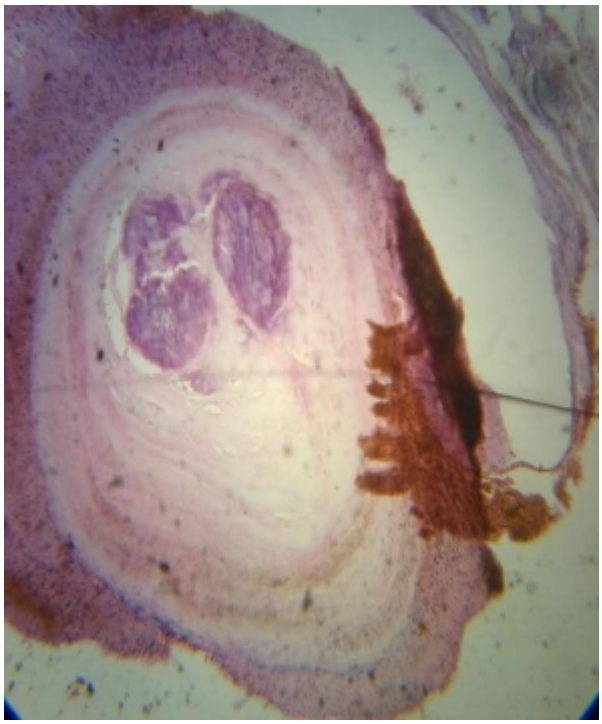

Fig 3: Calcified mass with in the vascular channel

Discussion:

It was in 1991 Calonje E and Fletcher CD described sinusoidal haemangioma(SH) as an unusual entity and considered it to be an adult variant of cavernous

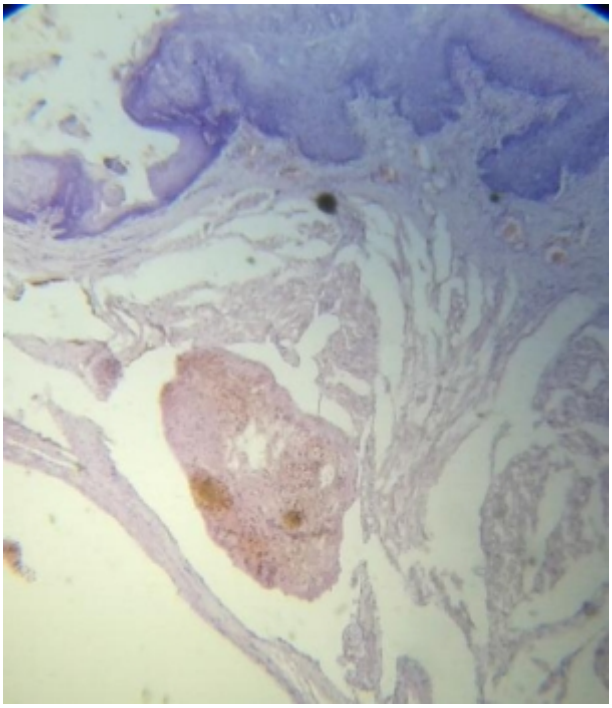

Fig 4: Pseudopapillary proliferation of the endothelium with thrombus formation

hemangioma.[5] Sinusoidal haemangiomas are rare in the oral region, the deceptive rarity of the reported cases may be due to despite having distinct features, it may be misdiagnosed as haemangioma or cavernous haemangioma. Pathogenesis of SH is unknown but it is assumed to be due to abnormalities in vasculogenesis and angiogenesis.

Sinusoidal haemangiomas presents as solitary, painless bluish subcutaneous nodules with a wide anatomic distribution.

They predominantly occur in extremities, trunk including mammary glands and scalp. They are relatively small tumors with slow growth seldom occur intramuscularly. Studies have also shown a female predilection of these lesions. Clinically, they appear as nodular lesions when they are located subcutaneously in the deep dermis.[5] However there are also reports of rare plurinodular polypoidal[6] forms as well as cystic structures that are covered by normal mucosa.

It is advisable to do diagnostic imaging of all the suspected vascular lesions before biopsy.[7] It include ultrasonography, magnetic resonance imaging (MRI), and computed tomography (CT). Although CT scan is the most accessible and used imaging technique, MRI being more efficient in the analysis of soft tissue lesions, it is more helpful in studying the lesions in tongue. CT imaging gives information regarding the composition, size and the extent of the lesion.

Aspiration cytology have limitations and were inconclusive in our case since the specimens obtained were found to contain only hematic material.

The histopathological features of Sinusoidal haemangiomas that overlap with various benign and malignant vascular neoplasms. Microscopic examination of the subcutaneous tissue displays lobular architecture of sinusoidal haemangioma. Thin walled inter communicating vascular channels lined by flattened endothelial cells. Stroma is scanty with back to back arrangement of dilated and congested interconnecting vessels with a prominent sinusoidal pattern. The tumours often show thrombosis or hyalinization of vascular channels that lead to intravascular papillary endothelial hyperplasia, resulting in calcification.[5] These lesions sometimes may exhibit dysplastic features like nuclear pleomorphism and hyperchromatism.

Cavernous haemangioma is considered as the most common the differential diagnosis of sinusoidal haemangioma since they have dilated vessels interspersed in subcutaneous tissue. However they differ from sinusoidal haemangioma in its large size, tendency to occur predominantly in childhood and its occurrence mainly in upper body. Histologically cavernous haemangioma shows a non-lobular, poorly demarcated structure. Pseudo-papillary structures are also absent.[6]

Occasional dysplastic features like pleomorphism and nuclear hyperchromatism may lead to misdiagnosis of angiosarcoma. But angiosarcoma is an interstitial lesion whereas the latter is a subcutaneous lesion.[5] Diffuse infiltration, atypical mitosis along with multi-layered endothelial cells are other features that help to distinguish between the lesions.

Subcutaneous haemangiomas such as subcutaneous pyogenic granuloma, spindle cell haemangioma are considered to be part of the differential diagnosis. Subcutaneous pyogenic granuloma also known as lobular capillary haemangioma is characterized by the presence of distinctive lobules of dilated and congested capillaries. The myxoid stroma surrounding the angiomatous tissue contains spindle and stellate shaped connective tissue cells with occasional mast cells in it.8 Spindle cell haemangioma has characteristic cavernous or slit like vascular proliferations that are frequently lined by a thin layer of endothelium with alternating

areas of spindle cells which aids in distinguishing it from sinusoidal haemangioma.[9] Present case had all the classical histomorphological features of sinusoidal haemangioma with some degenerative changes.

Immunohistochemical studies by Suurmeijer AJ et al. (2007) reveals, these lesions are positive for markers such as CD31, CD34 and negative for D2-40, confirming the endothelial vascular origin of these tumors. To differentiate angiosarcoma, immunohistochemical assessment of Ki67 index may also be performed.[10]

The cure of sinusoidal haemangioma is by local excision. Other treatment modalities of Haemangioma include embolization, corticosteroid treatment, excision, electrolysis, thermocautery, sclerotherapy, immunomodulatory therapy with interferon alfa-2a, and laser photocoagulation.[11]

Conclusion:

Sinusoidal haemangioma is an unusual variant of Cavernous haemangioma and its occurrence in the oral region is very rare. It is important to differentiate sinusoidal haemangioma from cavernous haemangioma and angiosarcoma. We report a case of sinusoidal haemangioma with typical histopathological presentation. An interdisciplinary approach is often necessary for diagnosing sinusoidal haemangioma. Thus, clinical characteristics, as well as imaging, histopathology and immunohistochemical investigations can lead to the diagnosis of sinusoidal haemangioma. Microsurgical therapy is the treatment of choice and involves an extensive excision of tumour formation in order to prevent relapses.

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