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**Hemangioma in the lower labial vestibule of an eleven-year-old girl: A case report**

Aloyouny AY *et al*. Hemangioma in the lower labial vestibule

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**Abstract**

BACKGROUND

Hemangioma is a vascular benign tumour of endothelial origin. It appears commonly in the first decade of life with increases incidence in females. Hemangioma is not common to happen in the oral cavity and it is extremely rare to appear in the labial vestibule.

CASE SUMMARY

We present a case of an 11-year-old girl who complained of a painful, slowly growing mass which was consistent with the capillary hemangioma in the left mandibular vestibule.Vascular tumor such as hemangioma in the mandibular vestibule is extremely rare; hence, the clinical definitive diagnosis is very challenging. Therefore, radiographic imaging and histopathologic analysis are crucial to reach to the final diagnosis for proper management.

CONCLUSION

Comprehensive clinical evaluation, proper diagnostic imaging and microscopic analysis of the mass establish a precise diagnosis of the hemangioma for better management.

**Key Words:** Capillary hemangioma; Vascular malformation; Labial vestibule; Childhood; Case report

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**Core Tip:** Although hemangioma rarely occurs in the oral cavity, it should be considered in the diagnosis of a red-bluish isolated mass. In this case report, the patient presented with a painful, slowly growing mass in the left labial vestibule which resulted in asymmetry and swelling of the lower lip. The final diagnosis of the mass was consistent with capillary hemangioma in the mandibular vestibule. Early detection and treatment of oral masses is essential to avoid any complications.

**INTRODUCTION**

Hemangioma commonly appears early in life with increases incidence in females more than males. It usually gets smaller with time until it completely disappears. Mulliken *et al*[1] (1982) published the novel classification of vascular lesionand the International Society for the Study of Vascular Anomalies (2018) has provided updated guidelines for vascular anomalies classification. Accordingly, the vascular anomalies have been classified into vascular malformations and vascular tumors. Vascular tumors include non-harmful, locally destructive, and malignant lesions, whereas vascular malformations include simple and combined malformations.

Hemangiomas are vascular benign tumours of endothelial origin, presenting clinically with varying sizes and shapes. In some cases, hemangioma could cause functional disability and disfiguring appearance, which may lead to psychological issues. Histologically, hemangioma has another classification, into either cavernous or capillary types.

Hemangioma is uncommon to occur in the oral cavity and it is extremely rare to appear in the labial vestibule. To our knowledge and based on the review of English literature (PubMed), this case is the first report of capillary hemangioma in the mandibular vestibule. In this report, we present an 11-year-old girl patient complaining of a slowly growing mass that caused labial swelling and asymmetry. The diagnosis was consistent with capillary hemangioma in the mandibular vestibule.

**CASE PRESENTATION**

***Chief complaints***

An 11-year-old healthy girl was referred to the oral and maxillofacial surgery clinic for evaluation of labial asymmetry and swelling.

***History of present illness***

The patient had a two-month-history of a slowly growing lesion in the left side of the lower labial vestibule accompanied with persistent mild pain. The patient and her parents reported no history of trauma at the site of the mass.

***History of past illness***

The patient was healthy and did not undergo any surgeries.

***Personal and family history***

The parents revealed no significant family history and no genetic abnormalities.

***Physical examination***

**Physical and systemic examination:** The patient was healthy and had only taken Tylenol 15 mg as needed for fever.

**Extraoral examination:** Extraoral examination showed lower lip asymmetry and swelling in the left side.

**Intraoral examination:** Intraoral examination showed a 2.0-1.5 cm, solitary, fluctuant, bluish, smooth, palpable submucosal mass, rubbery in consistency, tender, and blanch on pressure (positive diascopy test). The submucosal mass located in the left mandibular vestibule opposite to tooth number 32 and 33 (23 and 33, according to the FDI World Dental Federation Notation) (Figure 1).

***Imaging examinations***

A panoramic radiograph showed normal structures with no significant pathologic findings. Additionally, Color-Doppler-ultrasound was performed to confirm the nature of the lesion. The imaging interpretation revealed a slow-flow vascular lesion in the left lower vestibule and attached to the lower orbicularis oris muscle.

**FINAL DIAGNOSIS**

The final diagnosis was based on histopathological analysis. The histopathological diagnosis was consistent with capillary hemangioma in the labial vestibule (Figure 2).

**TREATMENT**

Complete surgical removal of the lesion to reduce the risk of the recurrence.

**OUTCOME AND FOLLOW-UP**

At two-, four- and eight-week-follow up, the site of the surgery healed well with no sign of bleeding, infection, and swelling. At one- and three-year- follow up, there was no recurrence of the lesion or complications noted. Additionally, the patient was in a good health.

**DISCUSSION**

In 1982, vascular anomalies were categorised into two main categories: Vascular malformations and vascular tumors. Hemangiomas are true neoplasms represented by increased rate and proliferation of endothelial cell turnover. On the other hand, vascular malformations are localised abnormality and disorganisation of the blood vessel caused by defects in vascular development[1-3]. Simple vascular malformations are classified histologically, based on the vessel size, into capillary, venous, lymphatics, arteriovenous fistula, and arteriovenous malformations. Vascular lesions are further categorised into non-harmful, locally destructive, and malignant lesions[4]. Namely, hemangioma is a neoplasm of endothelial origin which is commonly found in the early years of life and then the neoplasm regresses gradually with age[5]. Intraoral and intramuscular hemangiomas are rare, dissimilar to cutaneous and subcutaneous hemangiomas. Oral hemangiomas could occur in more than 6% of infants and have high prevalence in female presenting 3:1 (female:male). Infants are more likely to develop oral hemangiomas if they fall in one of the following conditions; infants who are born to older mothers, twins or triplets, premature, or have low birth weight[6]. Hemangioma is a common vascular benign tumor which falls under the category of benign vascular tumors and it is further divided into capillary and cavernous hemangioma[2].

Capillary hemangioma is a common lesion, but it rarely occurs in the oral cavity. According to Matsumoto *et al*[7], 45.2% of capillary hemangiomas occur on buccal mucosa, 35.5% on the tongue, and only a small percentage occur in the lip, gingiva and palate. Capillary hemangiomas are firm in consistency and have a limited history of symptoms. Although the exact cause of oral hemangioma is not fully understood, hormonal changes, embolic phenomenon and genetic mutations are believed to play an important role in the tumor development[8].

Hemangiomas are hypothesised to develop because of both angiogenesis and vasculogenesis through three different stages, as follows: Endothelial cell proliferation stage, rapid growth stage and spontaneous disappearance. Endothelial cell proliferation is stimulated by many factors, such as basic fibroblast growth factor, vascular endothelial growth factor and transforming growth factor-beta. Then, the quantity of endothelial cells is sustained, and each cell increases in size, leading to comprehensive enlargement of the structure size. At the end, spontaneous involution occurs when the endothelial cells are replaced by connective tissue, adipose, and fibroblast, and the number of small vessels decrease in quantity[9].

Hemangioma could be classified clinically as congenital or infantile (previously named strawberry or juvenile). Congenital hemangioma presents at birth and does not demonstrate proliferation stage. In contrast, infantile hemangioma may develop at the first months of the infant life and show a proliferative phase during the period of six to twelve months; then, most cases spontaneously regress between the age of six to nine years. High percentage of hemangiomas disappear completely in childhood, with < 20% carrying on to puberty[10,11]. Oral hemangioma presents as a solitary, soft, fluctuant, compressible, smooth, red, or bluish submucosal mass. Significant variations may present based on the depth and site of the mass. Superficial masses are easy to visualise and may present as pedunculated, sessile, or lobulated and reddish in colour. In contrast to deeper masses, they appear as a dark blue discolouration recognisable from surrounding normal colour mucosa. It also reveals tenderness on palpation and blanch on compression with glass slide (positive diascopy test)[12]. In this case, the differential diagnosis of the tumor was written down as vascular anomalies, including hemangioma, and vascular malformation, including venous, capillary, lymphatic and arterial malformations. Salivary gland tumor, mucocele and angioleiomyoma were also considered.

It is worth mentioning that vascular malformations, salivary gland tumor, mucocele and angioleiomyoma were all excluded because the lesion showed a slow-flow vascular lesion by using Colour-Doppler-ultrasound, which is highly consistent with hemangioma.

Histological analysis of vascular anomalies, including capillary hemangioma, is still the most acceptable and accurate method of diagnosis[3]. Microscopic description of capillary hemangioma illustrates several dilated capillaries lined by endothelial cells, filled with blood, and surrounded by inflammatory infiltrate.

Hemangioma is mostly characterised by its benign feature and has high tendency of involution by itself over time. However, sometimes hemangioma requires intervention, especially in case of impairment in breathing, swallowing and speech. The first line of evaluation and diagnosis would be by Color-Doppler ultrasound imaging. This imaging modality is non-invasive, cost-effective and has no risk of radiation. If intraosseous lesion is anticipated, other imaging modalities could be useful for the diagnosis, such as a contrast-enhanced magnetic resonance imaging (MRI), computed tomography (CT), and angiography[13]. A contrast-enhanced MRI and CT imaging identify the shape, size, and calcification of the tumor. The Color-Doppler ultrasound imaging modality was the suitable choice for the patient due to many factors, such as financial issues and the age of the patient[14].

Choosing a suitable method for managing hemangioma is based on multiple factors such as the aesthetic consideration, clinical nature, size, site, growth rate, accessibility, extent of the tumor, and age of the patient. Hemangioma could be managed by different ways; for instance, surgical excision of the tumor, embolization, electrosurgery, cryosurgery, laser, steroid injection, or sclerosing materials. In case of small oral hemangioma, the commonly used method is the total surgical excision of the whole mass to decrease the potential risk of recurrence[15]. However, if the lesion is large and located in a significant part of the mouth, such as the tongue, in this case the surgical excision of the lesion would not be preferred, so as to avoid post-surgical complications in swallowing and speech. Sclerotherapy is recommended to manage large hemangiomas in the oral cavity in which 3% sodium tetradecyl sulfate or ethanolamine oleate is injected into the main vessels of the lesion to destroy the endothelial cells, leading to lesion destruction. Precautionary measures should be taken to avoid bleeding during the surgical procedure or afterward during recovery phase. In the present case, the tumor was excised surgically with a thin rim of the attached orbicularis oris muscle to decrease the risk of recurrence[16,17].

**CONCLUSION**

Although hemangioma rarely appears in the oral cavity, it should be considered in the diagnosis of a red-bluish isolated mass. Comprehensive clinical evaluation, proper diagnostic imaging and microscopic analysis of the mass establish a precise diagnosis of the hemangioma for better treatment.

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**Footnotes**

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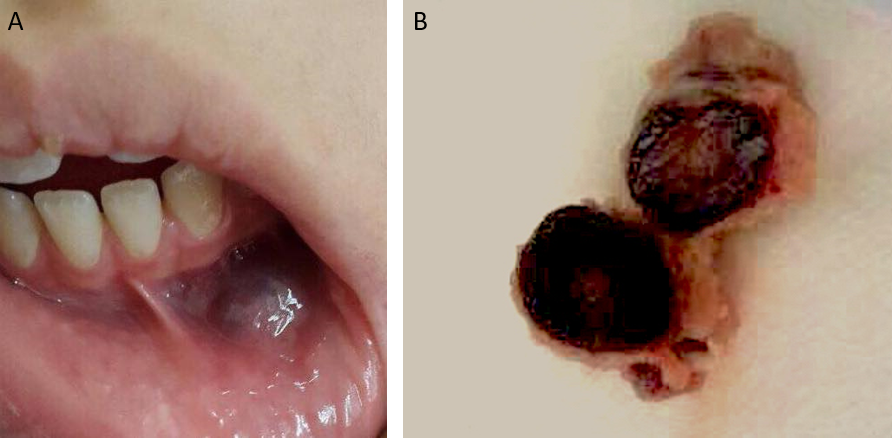
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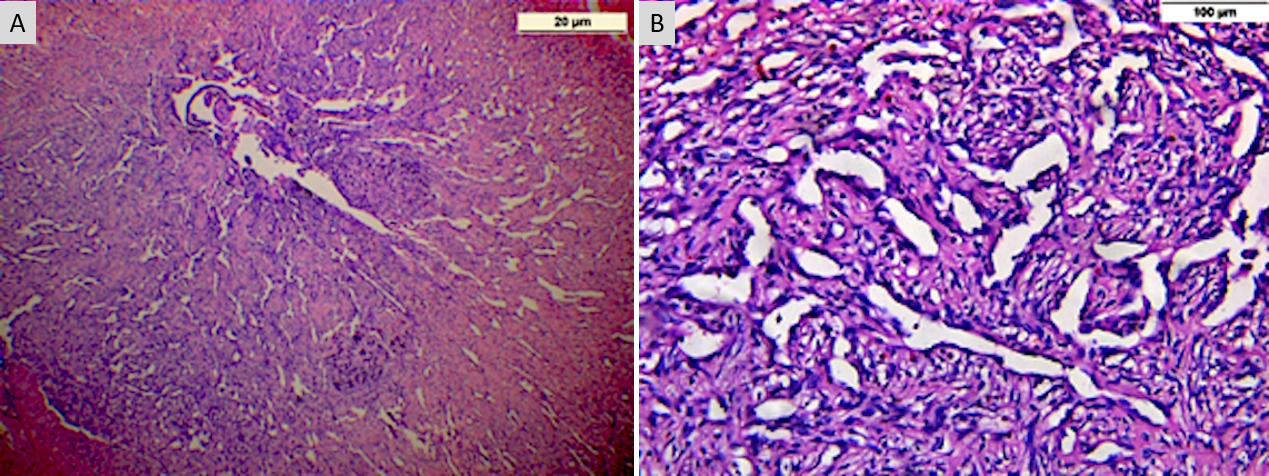
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**Figure Legends**



**Figure 1 Intraoral examination and gross examination of the surgical specimen.** A: Intraoral lesion measuring 2.0-1.5 cm, solitary, fluctuant, bluish, smooth, palpable submucosal mass located in the left mandibular vestibule; B: A single, solid mass, bluish in color, and rubbery in consistency.



**Figure 2 Histopathological analysis showed numerous thin-walled blood vessels with dilated blood vessel spaces, areas of hemorrhage and chronic inflammatory cell infiltrate which is consistent with capillary hemangioma in the labial vestibule.** A: 20 μm; B: 100 μm.



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