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Case Report

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Hemangioma of the Oral Cavity; Un Unusual Presentation

Ashwag Mohammed Alwagdani

Department of Otolaryngology, Division of Head and neck Surgery, King Fahad Armed Forces Hospital, Jeddah, Saudi Arabia

***Corresponding author**
Ashwag Mohammed Alwagdani, Department of Otolaryngology, Division of Head and neck Surgery, King Fahad Armed Forces Hospital, Jeddah , Saudi Arabia, E-mail: dr.wagdani_am@hotmail.com

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Abstract

Hemangioma is a developmental vascular malformation capillaries that do not involute but persist and grow very slowly over years. The diagnosis of hemangiomas is mainly based on clinical evaluation. They tend to occur in the oral cavity, on the tongue, buccal mucosa and labial mucosa. Isolated hemangiomas in the tonsillar tissue is a rare occurrence. In this we report a case of adult tonsillar hemangioma of left side associated with recurrent tonsillitis. She was effectively managed surgically with preoperative embolization.

Keywords: Hemangioma, Oral Cavity Hemangioma, Velopharyngeal Insufficiency

Case Report

A34–year–old female presented to our Out-Patients clinic in July 2019 with complaints of recurrent throat pain and fever for the past 9 months. She also gave history of constant foreign body sensation on left tonsillar region. Intra-oral examination revealed soft purplish mass was seen filling the left tonsillar region. On palpation, mass was soft, non-tender, not bleeding on touch. Neck examination revealed unremarkable. All other local and systemic examinations were normal. Routine blood, urine were normal. Computed tomography (CT) scan carotid angiogram of the neck showed faint vascular blush within the left tonsil and prominent vein noted connecting the left external jugular vein and internal jugular vein with a focal area of capillary blush noted medial to the lateral pterygoid muscle [Figure1&2]. Connection to external carotid artery branches and the relation of branches to the left tonsil was difficult to be identified in this study. Conventional angiogram with embolization for feeding vessel done 48hrs preoperative. Clinical and radiological diagnosis of chronic tonsillitis with left tonsillar hemangioma was made. Since patient was symptomatic tonsillectomy together with excision of hem angiomatous tissue was done under general anesthesia by bipolar diathermy. Postoperative patient discharge home in good condition. The Histopathological report confirmed cavernous hemangioma of left tonsil. Two weeks postoperative patient had symptoms of velopharyngeal insufficiency (VPI) include hyper nasality and Nasal regurgitation of liquids with swallow. Two months following surgery, the affected area had completely healed without intervention and there were no more complications [Figure3]. Patient is on regular follow up till now.

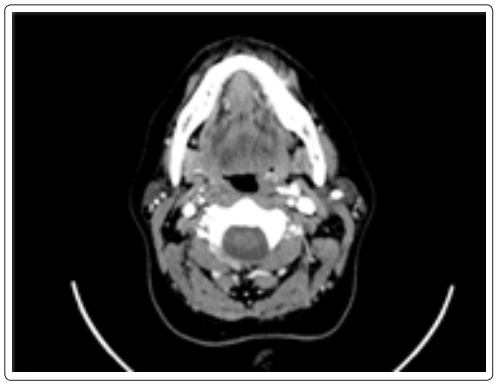

Figure1: Arterial supply from external carotid artery

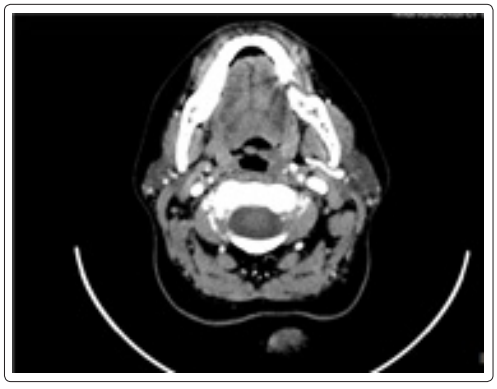

Figure 2: Venous supply from external jugular vein

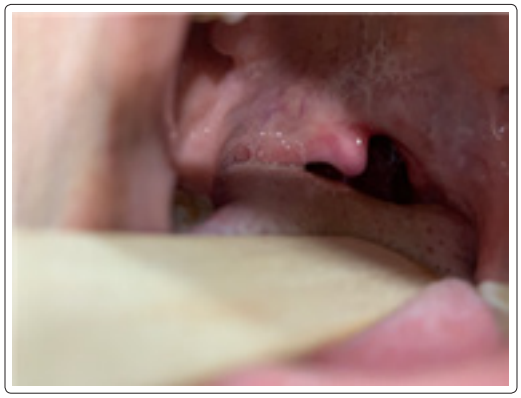

Figure 3: 2 months postoperative

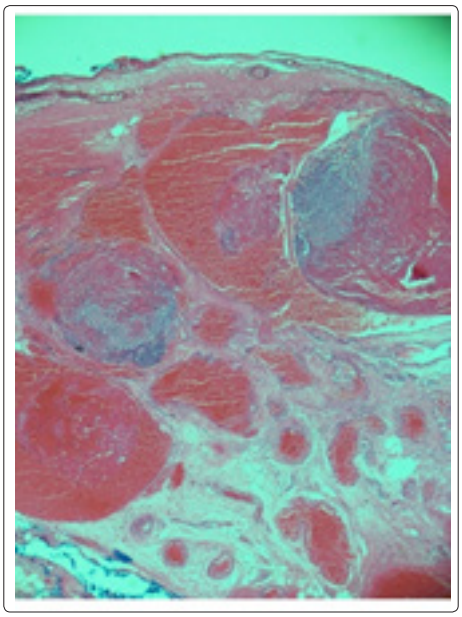

Figure 4: Numerous dilated vascular spaces lined by endothelial cells

Discussion

• A hemangioma is a benign lesion of blood vessels or vascular elements. The majority of oral and oropharyngeal hemangiomas seem to be of a developmental nature. In some instances lesions are probably a mixture of hemangioma and lymphangioma, leading to the term *angiomatosis* [1]. Vascular anomalies can present anywhere in the oral and oropharyngeal area such as in the tongue, lip, tongue, anterior gingival and buccal mucosa and very rare presentation in the tonsils. First case of hemangioma was documented by Liston (1843). Later in 1867, Virchow described the first case of vertebral hemangioma. Kasabach and Merrit (1940) reported a case of hemangioma involving the skin and deep soft tissues of the thigh that was associated with extensive purpura there after double eponym **Kasabach–Merritt syndrome** came to be used in as hemangioma with thrombocytopenia [3].

• The term "hemangioma" is misused for various vasculoformative tumors, for this confusion, the International Society for the Study of Vascular Anomalies has recently provided guidelines to differentiate these two conditions, according to the novel classification first published by Mulliken and Glowacki in 1982[2].The vast majority of vascular anomalies were divided into two main categories (a)vascular tumors: hemangioma (HEM), pyogenic granuloma, rapidly involuting congenital hemangioma, noninvoluting congenital hemangioma, hemangiopericytoma, tufted angioma and kaposiform hemangioendothelioma and (b) vascular malformation. Most cavernous hemangiomas of the head and neck region have recently been renamed as venous vascular malformations [4]. The major distinction between hemangiomas and the venous vascular malformations is that the latter do not involute and may actually grow with time, hormonal influences, infection, thromboses, or trauma [4]. Hemangiomas are a true tumor of endothelial cells with rapid growth until 6-8 months and involute by 5-9 years of age and histology there is pronounced endothelial turn over are relating into elevated fibroblast growth factor(FGF) signaling and they do happen to stain glucose transporter protein isoform 1 (GLUT1) positive. Instead at vascular malformations are not tumors but result from abnormal vascular or lymphatic vessel morphogenesis and they are presented at birth with normal endothelial turn over and FGF signaling.

The diagnosis of hemangioma is based on clinical history and physical examination. Imaging studies may be necessary to clarify and confirm the diagnosis, and in order to analyze the extent of the lesions by permitting an evaluation of their non-visible component as well as the affection of neighboring structures. The imaging techniques employed for hemangiomas include MRI, CT, and CT with contrast media, ultrasonography and angiographic techniques (arteriography, phlebography) [5, 6].

Syndromes associated with cavernous hemangiomas are Sturge Weber syndrome, Kasabach Merritt syndrome, PHACE (posterior fossa brain malformations, haemangioma of the face, arterial cerebrovascular anomalies, cardiovascular anomalies, eye anomalies, and sterna defects or supraumbilical raphe) syndrome [7].

The management of hemangiomas of the oral mucosa varies according to the age of the patient, the size of the lesion, the site of involvement and the clinical nature of the hemangioma. The range of treatment includes steroid therapy, carbon dioxide or argon laser therapy, sclerosing agents, surgical excision with or without ligation of vessels and embolization [8-10].

Conclusion

Hemangiomas are tumors identified by rapid endothelial cell proliferation in early infancy, followed by involution over time. All other abnormalities are malformations resulting from anomalous development of vascular plexuses. Tonsillar hemangioma is an uncommon form of oropharyngeal hemangiomas. Proper clinical diagnosis and preoperative investigation is essential for management of vascular tumors.

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