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Intramuscular hemangioma of masseter muscle: A report of unique case

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Case Report

Abstract

BACKGROUND AND AIM: Even though hemangiomas are prevalent tumors in the region of head and neck, they are comparatively rare inside the mouth and less frequently detected by dental professionals. Therefore, the aim of this case report is to present such a rare variation of hemangioma manifested within the substance of the masseter muscle.

CASE REPORT: We report a unique case of intramuscular hemangioma of masseter muscle in a 26-year-old woman complained of growth and swelling in the right cheek since 6-8 months before. The growth was surgical excised and diagnosed histopathologically as intramuscular mixed capillary with cavernous hemangioma.

CONCLUSION: Hemangiomas are rarely seen intramuscularly. This case presents an intramuscular hemangioma occurring within the masseter muscle. Early detection and management is required in order to avoid the potential complications associated with it.

KEYWORDS: Capillary; Cavernous; Hemangioma; Vascular Malformation; Benign Tumor

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Hemangioma (Greek: Haima: blood, angeion: vessel, oma: tumor) is defined as "a benign tumor of dilated blood vessels". Hemangioma of head and neck usually occur following birth showing swift proliferative phase, and then resolves completely by involution. It is also named as "strawberry hemangioma", and "Salmon patch" based on its location. They are never encapsulated, usually manifested as hyperplasia of capillaries and veins in the connective tissue.[1,2] They may be cutaneous (at sites like skin, lips, and deeper structures), mucosal (lining of the oral cavity), intramuscular (within the masticator and perioral muscles), or intra-osseous (within the mandible and/or maxilla).[3] Oral hemangiomas are rarely seen on the gingiva and periodontium at interdental gingival papilla, and spread laterally to involve adjacent teeth.[4] Even less frequently other sites like buccal and labial mucosa, lips, tongue, and palate are involved.[5]

Clinically, hemangiomas manifest as a soft mass of varying sizes; which may be smooth or lobulated, sessile or pedunculated. On visual examination, the lesion appears to be either red, pink, or purple, and it blanches on the application of external pressure. Hemorrhage may even occur spontaneously without external traumatic factor, or even after minimal trauma.[4] They are generally painless, but might functionally interfere with mastication.[3,4] While the superficial hemangiomas manifest as lobulated lesions showing blanching when finger pressure is applied, deeper lesions appear as dome-

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shaped with color ranging from normal to blue, and rarely blanch on pressure application.[6]

This report describes a unique case of intramuscular hemangioma of masseter muscle in an adult woman in the right cheek area.

Case Report

A woman aged 26 years, reported to a private dental college with a chief complaint of a growth and swelling in the right cheek since 6-8 months before. The patient had mild pain and discomfort while eating due to obstruction of occlusal area by the growth during mastication. Medical history and family history was noncontributory. Extra-orally, no changes were noticed. A comprehensive intraoral examination revealed well circumscribed, non-fluctuant swelling on the right cheek. The surface of the buccal mucosa was bright red with no surface ulceration (Figure 1). A radiographic diagnosis of desmoid tumor was made following magnetic resonance imaging. The lesion was surgically excised, and sent for histopathological investigation. The surgeon encountered profuse bleeding while excising the lesion.

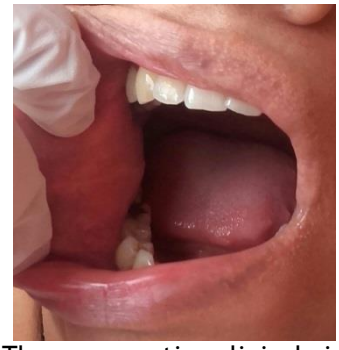

Figure 1. The preoperative clinical view showing a well circumscribed, non-fluctuant swelling on the right cheek

On gross examination, the biopsy specimen was brownish yellow in color measuring 6 × 4 × 2 cm, firm in consistency with adipose tissue attached to it. Sectioning of the gross specimen showed irregular areas of yellow and reddish brown discoloration (Figure 2).

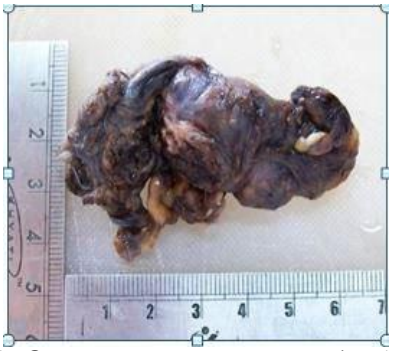

Figure 2. Gross specimen measuring 6 × 4 × 2 cm, reddish brown in color with adipose tissue attached to it

Histopathological examination using Hematoxylin and Eosin staining revealed the presence of numerous blood vessels of different sizes, with dense stroma along with longitudinal and transverse section of skeletal muscle fibers, areas of hemorrhage, and adipose tissue along with very mild inflammatory infiltrate (Figure 3).

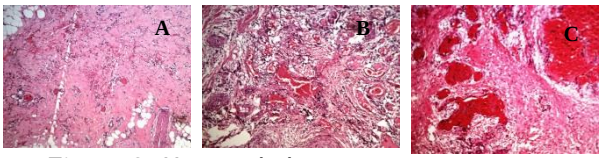

Figure 3. Histopathologic examination using Hematoxylin and Eosin staining, × 4 (A), × 10 (B), and × 10 (C), showing both capillary and cavernous components with presence of numerous blood vessels of different sizes, dense stroma, longitudinal and transverse section of skeletal muscle fibres, areas of hemorrhage, and adipose tissue along with very mild inflammatory infiltration

Prominent endothelial cells were seen lining the capillaries of various sizes along with extravasated red blood cells (RBCs). Marked proliferation of endothelial cells were also observed. Very few plasma cells and lymphocytes could be seen scattered throughout stroma. Some of the medium sized vessels showed presence of organizing fibrin thrombi (Figure 4). The histopathologic diagnosis of intramuscular mixed capillary and cavernous hemangioma (venous hemangioma) was made. Further follow up through telephonic conversation with the

patient was done, and the healing was reported as uneventful.

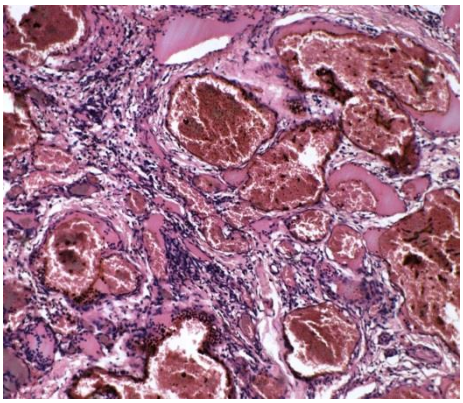

Figure 4. Histopathologic examination using Hematoxylin and Eosin staining (× 40), showing capillaries with red blood cells (RBCs) along with sparse lymphocytes and plasma cells scattered throughout stroma

Discussion

Vascular lesions are usually classified as either hemangiomas or vascular malformations.[7] Difference between arteriovenous (AV) malformation and hemangioma is that, AV malformations are structural anomalies of blood vessels present at birth, and persist throughout life, showing normal endothelial cell growth which affects the capillaries, veins, or lymphatics. They are more stable, fail to regress, and often shows signs of beating, and thrilling, and lastly do not involute. Whereas, hemangiomas are tumor-like malformations showing marked epithelial cell proliferation along with disorganized masses of endothelium-lined vessels that are filled with blood. They exhibit a rapid growth phase, followed by an involuting phase.[4,8,9]

Mulliken and Glowacki[8] elicited a most accepted classification scheme which divided the vasoformative tumors into 2 broad groups, hemangiomas and vascular malformations along with old and new nomenclatures (Table 1).

Hemangiomas involve the head and neck region in majority, and are frequently seen in whites than other racial groups.[4] Women are more affected than men,[10,11] as seen in our case. In younger children, the proliferative phase of hemangioma usually lasts for 6 to 10 months, after which the tumors slow in growth and begins to involute. By the age of 5 years, most of the red color disappears, and about 50% of all hemangiomas will show complete resolution by 5 years of age, with 90% resolving by age of 9 years.[6] Occasionally, older individuals are affected, as in our case.[11,12]

Table 1. Classification of vasoformative tumors by Mulliken and Glowacki[8]

Vasoformative tumor	New nomenclature	Old nomenclature
Hemangiomas	Capillary hemangioma	Strawberry hemangioma
	Cavernous hemangioma	Juvenile hemangioma
	Mixed hemangioma	Parotid hemangioma
Vascular malformations	Venous malformation	Cavernous hemangioma
		Hemangiomatosis
	Intramuscular venous malformation	Intramuscular hemangioma
	Capillary malformation	Capillary hemangioma
		Port-wine stain
	AV malformation	AV hemangioma
		Arterial angioma
		AV aneurysm
		Cirsoidangioma
		Red angioma
		Serpentine aneurysm
	Lymphatic malformation	Capillary lymphangioma
		Cavernous lymphangioma
		Lymphangioma
		Cystic hygroma

AV: Arteriovenous

Based on the histopathological appearance, hemangiomas are classified into two main type of cavernous and capillary.[9,10] Cavernous hemangiomas are comprised of thin-walled sinusoids or vessels which are large, along the uni-layered endothelium, and the thin septa of connective tissues separates them. On the other hand, capillary hemangiomas have numerous tiny capillaries lined by a uni-layered endothelial cells which is supported by a connective tissue stroma. Rarely, hemangiomas would show large as well as small capillaries, and are called as 'mixed hemangiomas'.[4]

A special type of hemangioma involving the skeletal muscle are noted in the region of head and neck, and are called intramuscular hemangioma which comprises only 0.8% of all hemangiomas. In the head and neck area, intramuscular hemangiomas are most frequently seen in the masseter muscle followed by the trapezius and sternocleidomastoid muscles. Histologically, they are seen as large and small proliferating vessels which are embedded within muscle tissue in the deep layer. They therefore have somewhat different characters from other types of hemangiomas. IHMs are usually seen in the first three decades of life, and not noticed until there is pain and enlargement. Etiological factors include hormonal change, infection, or trauma, as seen in this case.[13-15]

Differential diagnosis of intramuscular hemangioma should include masseteric hypertrophy, lymphangiomas, schwannomas, rhabdomyosarcomas, salivary neoplasms, telangiectasia, angiosarcoma, and other vascular appearing lesions of face.[14,3]

No intervention is required in the management of true hemangioma as it resolves by itself. However, 10%-20% may require intervention because of functional compromise, behavior, stages of growth, or regeneration, and the most important factors are the size and location. Horizon of treatment includes intralesional injection of fibrosing agent, electrocoagulation, flash lamp pulsed laser, interferon alpha-2b, and surgery.[2] In our case, surgical approach was preferred considered on the basis of size, location, and difficulty in swallowing.

Conclusion

Hemangioma of the oral cavity is of clinical importance, as they have a benign origin and behavior. Among the different types of hemangiomas, intramuscular hemangiomas seen in the buccal mucosa are relatively rare, and might mimic other lesions clinically and histologically. Dental surgeons must be aware of these kind of lesions and potential complication when excising such kind of lesions, as it may result in serious bleeding. Hence, the planning of the treatment modality should be done based on the diagnosis of the vascular lesions and their prognosis.

Conflict of Interests

Authors have no conflict of interest.

Acknowledgments

None.

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