

CASE REPORT

Cavernous Hemangioma of the Tongue

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ABSTRACT

Background: Vascular anomalies are divided into vascular tumors, hemangiomas being the most common, and vascular malformations. Most vascular anomalies are noticed at birth or occur during infancy, and generally involve skin or subcutaneous soft tissues. Adult onset hemangiomas are rare, and intramuscular location is extremely rare. Surgical excision is recommended for hemangiomas in adults, if they are symptomatic, or manifest growth.

Materials and methods: We report a rare case of a 51-year-old woman, with an intramuscular hemangioma of the tongue, presenting as a submental mass. Preoperative imaging for assessment of tumor extent was followed by a successful surgical excision.

Results: Postoperative course was uneventful with primary healing of the wound, and with no functional deficit of tongue function.

Conclusion: Although a variety of treatment approaches are reported for childhood hemangiomas, surgical excision is the preferred treatment for adult onset symptomatic hemangiomas. Preoperative work up should include imaging preferably with contrast enhanced magnetic resonance imaging (MRI). Embolization may be considered for larger lesions. Intraoperative hypotension should be avoided to ensure identification of the entire lesion to ensure complete excision.

Keywords: Hemangiomas, Surgery, Vascular.

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INTRODUCTION

Neoplasms (hemangiomas) and malformations (arterial, venous or lymphatic) of vascular origin occur most commonly in the head and neck region. The majority of these lesions involve the skin or subcutaneous soft tissues. Inter, and intramuscular lesions are rare. Many of

these lesions are congenital, or appear during infancy and show spontaneous regression as the child grows. On the other hand, adult onset hemangiomas are extremely rare. Dilemmas in accurate diagnosis and management occur due to unfamiliarity with the nature and pathophysiology of these lesions. Extensive overlap in clinical and histologic terminologies makes accurate diagnostic and therapeutic decisions difficult.¹

One commonly accepted classification proposed by Mulliken and Glowacki² defines two fundamental categories: vascular tumors (hemangiomas) and vascular malformations. The Hamburg classification system adopted an embryologic perspective to further aid in the classification of vascular malformations. It subdivides malformations into either an extratruncular or truncular form, based on the time of developmental arrest during embryonic life.³ Vascular malformations are structural anomalies, or errors of vascular morphogenesis. They are, therefore, often divided into 'slow-flow' (capillary, venous, lymphatic, or combined forms) or 'fast-flow' (arteriovenous fistulas and arteriovenous malformation).²

Hemangiomas on the other hand are true neoplasms of vascular origin. They present in two varieties, capillary and cavernous. Most small lesions are asymptomatic, and present only cosmetic concerns. On the other hand, larger lesions causing symptoms due to location, proximity to vital structures, impairment of function, or bleeding due to ulceration or trauma require surgical excision. Growth of the lesion is also another indication for consideration of surgical intervention.

We present here a rare case of intramuscular hemangioma of the tongue, presenting as a submental mass.

CASE REPORT

A 51-year-old female presented with the history of an enlarging submental mass since the spring of 2014. She sought consultation from her primary care physician who recommended an ultrasound of the neck, which showed a heterogenous solid submental mass measuring 3.2 × 2.3 × 2.9 cm of undetermined etiology. A fine needle aspiration biopsy was performed which showed a spindle cell lesion possibly of vascular origin. At that point, she sought consultation from a surgeon, who attempted excision of the submental mass, but found 'no tumor on exploration'. Some benign appearing fibroadipose tissue and a lymph node were excised. Pathology report

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of the excised tissue did not reveal any vascular lesion. Postoperatively, the patient continued to complain of a persistent mass, and therefore, a noncontrast computed tomography (CT) scan of the neck was obtained 1 month after the surgery, which was reported to show the absence of left submandibular gland and scattered bland-appearing lymph nodes with no pathologic appearing adenopathy. However, the patient noted that the original submental mass was still present and, in fact, became larger by the beginning of 2015. Therefore, a repeat CT scan with contrast was obtained and compared to previous CT which was reported to show persistence and growth of a mass measuring 3×2.5 cm in the floor of the mouth. The radiological interpretation was favoring a mass of vascular origin. At that point, the patient was seen by us in consultation for further treatment. Her past history is remarkable in that she had undergone excision of dermatofibrosarcoma of the anterior chest wall in 1987 and 1988. She, however, remains free of disease at that site.

On physical examination, the skin of her face and neck was unremarkable except for a well-healed scar from her previous surgical procedure in the submental region (Fig. 1). Diffuse swelling of the entire submental region beneath the scar was visible. On palpation, the submental mass was felt to be spongy and easily compressible, giving the clinical impression of a lesion of vascular origin, such as a hemangioma or lymphangioma.

Intraorally, there was no significant mucosal pathology. Further work up of the lesion included a magnetic resonance imaging (MRI), which clearly showed a well-demarcated, irregular lesion in the submental region extending into the deep musculature of the floor of the mouth between the digastric muscles and deep to the mylohyoid muscle. The lesion showed contrast enhancement, and was bright on T2-weighted sequence, confirming the diagnosis of a vascular lesion (Fig. 2).

Surgical excision was performed under general anesthesia. During the whole procedure, we maintained her systolic blood pressure over 100 mm Hg. This is important, since if the patient has hypotension, often the lesion decompresses spontaneously and is difficult to identify within the muscles, and its true extent is not appreciated leading to incomplete excision. After the incision was made through platysma, the anterior bellies of the bilateral digastric muscles were retracted laterally. At this point, the mass could be seen protruding from beneath the mylohyoid muscle. The mylohyoid was incised in the midline. The mass was displacing but not infiltrating the fibers of the geniohyoid muscle, which was dissected free. Thus, dissection was complex because of the intramuscular nature of the tumor. However, with careful dissection around the pseudocapsule of the tumor it could

be separated from the surrounding muscles (Fig. 3). The mass was removed in its entirety without any significant blood loss (Fig. 4). The surgical specimen showed a cavernous hemangioma with exuberant histocytic/myofibroblastic proliferation and histological features of Masson's tumor (intravascular papillary endothelial hyperplasia). By immunohistochemistry, the histocytic/myofibroblastic proliferation was positive for CD 68 and negative for CD 31 and ERG. These histological characteristics led to a diagnosis of cavernous hemangioma.



Fig. 1: Well-healed scar from previous surgical procedure in the submental region with a diffuse swelling of the submental region beneath the scar

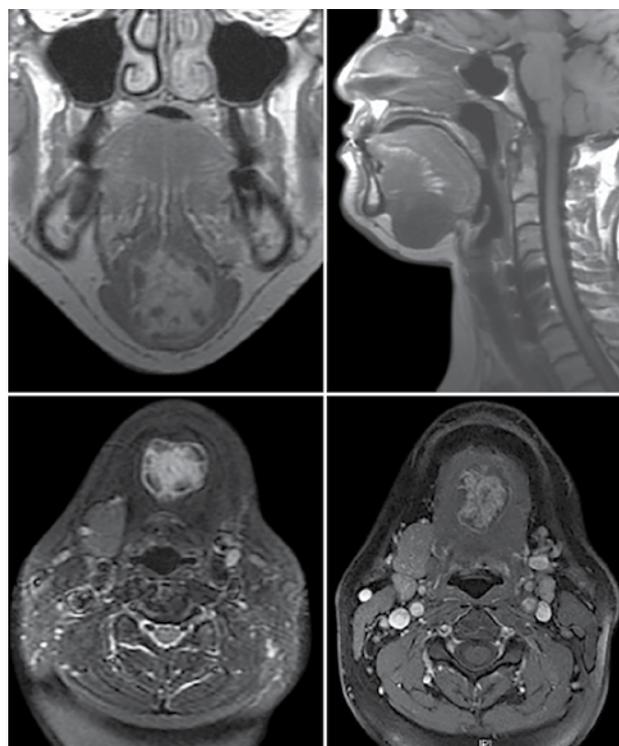


Fig. 2: Preoperative MRI scans of the neck. Clockwise from top left: coronal T1-post gadolinium; sagittal T1 without gadolinium; axial T1-post gadolinium; axial T2. The images clearly show a well-demarcated, vascular lesion in the deep musculature of the floor of the mouth between the digastric muscles and deep to the mylohyoid muscle. The T2 bright lesion confirms the diagnosis of a vascular lesion

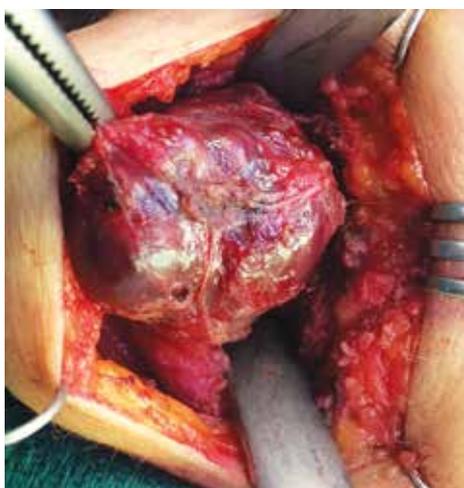


Fig. 3: Mass protruding out from the geniohyoid muscle, deep to the mylohyoid muscle

Postoperative course was uneventful with primary healing of the wound, and with no functional deficit of the tongue.

DISCUSSION

Mass lesions of the submental space represent a diverse group of histopathologic entities. The differential diagnosis should include reactive lymphadenitis, lymphoma, metastatic lymph nodes, dermoid cyst, abscess, and benign neoplasms, such as lipoma, hemangioma or tumors of neuromuscular origin, or their malignant counterparts.⁴ Radiologic imaging, such as CT or MRI and fine needle aspiration biopsy are essential preoperative tests to arrive at an accurate diagnosis.

Adult onset hemangiomas can occur anywhere, but are most often seen in the head and neck region. Intramuscular hemangiomas are rare. In the head and neck, the most common site for intramuscular hemangioma is the masseter muscle.⁵ The diagnosis of intramuscular hemangioma in other locations can be difficult. In most instances, accurate diagnosis of a hemangioma or vascular malformation requires assessment of both clinical findings and diagnostic imaging. Ultrasonography, MRI, CT, and conventional angiography each have a role in assisting diagnosis, but CT and MRI are optimal firstline diagnostic tools. These imaging modalities also play a central role in defining the extent of the lesion for appropriate treatment and assessing treatment efficacy.⁶

Most small asymptomatic hemangiomas can be left alone under observation. On the other hand, large or symptomatic lesions causing impairment of function or



Fig. 4: Surgical specimen showing complete excision of the tumor

esthetic deformity require surgical resection. For very large lesions, selective angiography and embolization may be considered to minimize blood loss. However, well-defined lesions of up to 4 to 5 cm do not need embolization and can be safely excised without significant blood loss.

Intraoperative hypotension should be avoided, since it often causes decompression of the lesion, and makes its identification and extent extremely difficult to define. We prefer to keep the systolic pressure above 100 mm Hg. Meticulous and slow dissection through the muscle fibers, and the pseudocapsule of the tumor, allows a bloodless and complete excision. The lesion is cured, if completely excised.

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