CASE REPORT

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GLOSSAL ANGIOMYOMA: IMAGING FINDINGS AND ENDOVASCULAR TREATMENT

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Abstract: Background. An angiomyoma is an uncommon, benign tumor characterized by numerous vascular channels intermixed with bundles of smooth muscle cells. Oral manifestations are quite rare. We describe for the first time the CT, MRI, and angiographic imaging features and successful preoperative endovascular embolization of an angiomyoma of the tongue. The pathologic findings before and after embolization are also described.

Methods and Results. A 25-year-old man was seen with a rapidly enlarging tongue mass. Imaging studies revealed the extent and hypervascular nature of this tumor. The diagnosis of angiomyoma was confirmed by histologic examination. Preoperative embolization proved to be helpful in the surgical management of this lesion.

Conclusions. Angiomyoma should be considered in the differential diagnosis of any well-circumscribed, hypervascular, soft tissue tumor in the mouth. In addition, endovascular embolization may be a useful adjunct that facilitates resection. © 2004 Wiley Periodicals, Inc. Head Neck 26: 1084–1088, 2004

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Angiomyomas, sometimes called angioleiomyomas or vascular leiomyomas, are benign, soft tissue neoplasms composed of encapsulated bundles of smooth muscle cells and prominent vascular channels. These tumors can be classified into solid, venous, and cavernous subtypes on the basis of their cytoarchitecture. Most descriptions of this rare tumor were written in the pre-MRI era and focused on the histologic findings, surgical management, and demographic characteristics of this patient population. To the best of our knowledge, approximately 10 cases involving the tongue have been reported, and none have discussed the cross-sectional imaging findings or nonsurgical adjuncts to treatment. We describe for the first time the CT, MR, and angiographic imaging features and successful preoperative endovascular embolization of an angiomyoma within the oral cavity.

CASE REPORT

A 25-year-old man was initially seen with posterior tongue irritation and a rapidly growing mass involving the left posterior oral tongue and tongue base.
He underwent two biopsies of this mass 10 days apart, each time bleeding vigorously, necessitating emergent intubations. Blood loss associated with the second biopsy was estimated to be 500 mL. Because of his symptoms, the patient elected to have the mass resected and a tracheostomy performed in anticipation of surgery. Preoperative CT, MRI and magnetic resonance angiography (MRI/MRA), and catheter angiography, followed by selective endovascular embolization of the mass, were subsequently performed.

A CT scan (Lightspeed-16; GE Medical Systems, Waukesha, WI) demonstrated a well-circumscribed mass of the left posterior tongue and tongue base with mildly heterogeneous enhancement (Figure 1). An MRI (Signa 1.5-T MR Scanner, GE Medical Systems) further characterized this mass by its isointense signal on T1-weighted sequences, hyperintense signal on T2-weighted sequences, and intense enhancement with gadodiamide contrast (Omniscan 0.1 mmol/kg; Amersham Health AS, Oslo, Norway). There was compression of the airway at the level of the oropharynx with lateral displacement of the constrictor pharyngeal muscles. The lesion abutted the soft palate and base of the tongue without evidence of extension into the parapharyngeal fatty areolar tissues (Figure 2). An MRA of the neck with two-dimensional time-of-flight, phase-contrast, and three-dimensional multiple overlapping thin-slab acquisition (MOTSA) techniques demonstrated no definite increase in vascularity.

A markedly hypervascular mass of the left tongue base, extending to the midline at its posterior margin, was seen on catheter angiography. It derived its arterial supply solely from branch vessels of the left external carotid artery. Superselective catheterization showed two large lingual artery branches supplying the mass, including a dominant, purely tumoral branch originating from the more proximal lingual artery, as well as smaller branches originating from the remaining lingual trunk and proximal facial artery (Figure 3).

After diagnostic imaging, the patient underwent endovascular embolization under general anesthesia with systemic heparinization. A 5 F guide catheter was positioned in the left external carotid artery. A Rapid Transit microcatheter (Cordis, Miami, FL) was then coaxially advanced.

**FIGURE 1.** An axial CT image shows a well-demarcated mass (arrow) of the posterior tongue and tongue base with mildly heterogeneous enhancement.

**FIGURE 2.** Axial MR images depict a well-demarcated mass (arrows) characterized by signal that is (A) hyperintense on the T2-weighted fast spin echo sequence (TR/TE = 5800/102.7) and (B) isointense to muscle on the T1-weighted spin echo sequence (TR/TE = 716.7/14). Postcontrast axial (C) and coronal (D) T1-weighted images demonstrate intense enhancement within the lesion (TR/TE = 716.7/14 and 633.3/14, respectively). (On the coronal image, the curvilinear region of hypointense signal within the tongue mass, extending centrally from the superior surface, represents hemorrhage from the biopsy 4 days earlier.)
over a Headliner-16 microguidewire (Terumo, Tokyo, Japan) to sequentially select the tumor-feeding branches of the left lingual and proximal facial arteries. A slurry of polyvinyl alcohol (PVA) particles (TruFill; Cordis, Miami, FL) of 150 to 250 \( \mu \text{m} \) was used to embolize the dominant left lingual and proximal facial branches, whereas a slurry of 355- to 500-\( \mu \text{m} \) particles was used for the remaining left lingual artery branches that supplied the tumor (Figure 4).

One day after embolization, the patient underwent a left hemiglossectomy with resection of the mass. Blood loss was estimated to be only 30 mL. Gross pathologic examination of the tongue specimen demonstrated an elevated mass measuring 3.2 × 3.0 × 1.2 cm located laterally and posteriorly, with an indurated area measuring 1.8 × 1.0 cm anterior and medial to the main mass. Sectioning of the mass revealed a

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homogeneous, tan-brown, slightly lobulated, rubbery surface. Focally dilated vessels were seen, but central necrosis was not. Microscopy findings were consistent with a venous type of angiomyoma, with thick- and thin-walled vessels associated with bundles of uniform-appearing smooth muscle cells (Figure 5). The endothelial cells along the walls of the prominent vascular channels were immunoreactive to CD 31 and 34. The spindle-shaped stromal cells were immunoreactive to smooth muscle actin. The postembolization specimen confirmed the presence of PVA occluding numerous large vessels, in addition to the interval collapse of many of the surrounding vascular channels compared with the pre-embolization biopsy.

DISCUSSION

Leiomyomas are benign neoplasms of smooth muscle that are categorized as solid, vascular, or epithelioid (leiomyoblastoma). The solid leiomyoma is the typical form, occurring most commonly in the uterus or gastrointestinal tract.2 The vascular leiomyoma, also known as an angiomyoma or angioleiomyoma, is the next most common subtype, occurring most commonly in the subcutaneous tissues of the extremities. These are generally painful. Leiomyomas of any type are very rare in the oral cavity, making up only five cases in a series of 7748 involving all sites in one study.2 The most common subtype in the oral cavity is the angiomyoma, making up approximately two thirds of all variants.3 A recent review of 109 cases of oral angiomyomas in the literature found that nearly half involved the lip, whereas only 9% involved the tongue.4 Oral angiomyomas have been reported in patients from 3 to 85 years of age, with a mean in the early fifth decade. At this site, angiomyomas are most often slow-growing and painless.4,5 Regardless of location, the treatment is usually surgical, and recurrence is rare.

When a patient is seen with a focal tongue lesion, diagnoses to consider include abscess, foreign body, lingual thyroid, hemangioma, lymphangioma, cystic hygroma, retention cyst, and papilloma. Benign tumors to consider include fibromas, neurofibromas, granular cell tumor (myoblastoma), and minor salivary gland tumors such as pleomorphic adenoma. The most common malignant tumor of the tongue is squamous cell carcinoma.

Imaging plays a key role in the evaluation of composition, extent, and complications of any head and neck mass. In this case, the tongue mass enhanced intensely on both CT and MRI, suggesting marked hypervascularity. On the basis of clinical and radiologic findings, a lymphangioma, hemangiomia, or other vascular malformation was suspected. Although not seen in this case, the presence of phleboliths on CT would have made the diagnosis of hemangiomia or venous malformation more likely. MRI features of angiomyomas outside the oral cavity have been described as well-demarcated, strongly enhancing masses with an isointense to slightly hyperintense signal compared with muscle on T1-weighted images and hyperintense signal on T2-weighted images.6–10 We suggest that angiomyoma be considered in the differential diagnosis of any well-circumscribed, hypervascular mass of the tongue as well.

Angiography can be helpful in surgical planning by demonstrating the feeding and draining vessels and in defining the high-flow or low-flow hemodynamics of vascular lesions. In this case, angiography confirmed the hypervascular nature and further delineated the arterial supply not shown by MRA that proved important for surgical planning.

This case also showed the benefit of preoperative embolization of an angiomyoma of the tongue, in this instance with PVA. PVA is a nonabsorbable, particulate, embolic agent that comes in a variety of sizes from 45 to 2000 μm. It is commonly used for the preoperative embolization of hypervascular tumors of the head and neck, including juvenile nasal angiofibroma, glomus tumors, meningoia, and squamous cell carcinoma.11 Only a single report has described the preoperative embolization of an angiomyoma.6 In that case, the angiomyoma occurred in the nasal cavity, supplied by branches of the internal maxillary artery, and embolization was undertaken with microfibrillar collagen (Avitene), an absorbable embolic agent of 75- to 150-μm particle size. Potential advantages of the use of PVA over microfibrillar collagen include more flexibility in choosing particle size, depending on vessel size, target tissue, and hemodynamic state, and greater durability in the event that surgery is delayed or incomplete. In this case, the initial, small biopsy specimens produced excessive blood loss, requiring intubation for airway protection. After embolization of the dominant tumor-feeding vessels, blood loss at surgery was minimal. Correlation with the microscopy of the preembolization and post-embolization specimens demonstrates the interval occlusion of large vas-
cular channels by PVA and secondary collapse of the remaining channels, corresponding to significantly diminished blood flow.

In conclusion, we report the CT, MR, and angiographic findings of an angiomyoma of the tongue for the first time and discuss the added value of preoperative endovascular embolization for this lesion. We also suggest that any hypervascular mass within the oral cavity undergo angiography for evaluation for potential preoperative embolization to facilitate a safer, less complicated surgical resection.

REFERENCES