

Harmonic Scalpel Surgical Treatment of the Tongue Angioleiomyoma – Case Report and Review of the Literature

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ABSTRACT

Angioleiomyoma is benign soft tissue tumor composed of smooth muscle cells and vascular endothelium, characterized with slow growth, unspecific symptoms and rare malignant transformation. It is very rare in the head and neck region and complete surgical excision is the gold standard for diagnosis and treatment. We present a very rare case of angioleiomyoma of the tongue base treated with partial glossectomy with harmonic scalpel which shortened surgical procedure, reduced bleeding and postoperative complications.

Key words: angioleiomyoma, tongue, harmonic scalpel

Introduction

Leiomyoma is benign soft tissue tumor composed of smooth muscle cells that usually arise in gastrointestinal system and uterus, while it is very rare in head and neck area. Leiomyomas have been classified as solid, angioleiomyoma (vascular leiomyoma), and epithelioid variants (leiomyoblastoma). Angiomyomas or angioleiomyomas or vascular leiomyomas can be capilar (solid), venular or cavernous¹.

Most commonly angioleiomyoma occurs in the skin of lower extremities (59%) and upper extremities (15%). In the head and neck region there are 13% of angioleiomyoma that can be found in the skin of the nose or ears, nasal or oropharyngeal mucosa deep neck spaces like parotid region². Only 0,06% of all smooth muscle cell tumors arise in the oral cavity, whereas proportion of angioleiomyoma subtype 0,016%. The most common localization in the oral cavity is upper lip, than lower lip, hard and soft palate and cheek mucosa^{3,4}. Glossal angioleiomyoma is extremely rare and usually arises from tunica media of the blood vessels or from tongue mucosa.

Clinical feature and radiological findings are unspecific and differential diagnosis should include haemangioma, lymphangioma or other vascular malformations.

Partial glossectomy and complete surgical resection of the tumor is the golden standard in surgical treatment. It is very important to obtain good bleeding control due to a very rich blood supply of the tongue base and the tumor. Sometimes oropharyngeal angioleiomyomas can be hypervascular tumors and endovascular embolization may be a useful adjunct that facilitates resection⁵. Most of the glossal tumors can be resected with harmonic scalpel safely and without complications.

Ultracision technology that simultaneously combines resection and coagulation with minimal tissue injury is widely used in gynecology and general surgery and in the last ten years it has been used in head and neck surgery⁶.

We present a very rare case of angioleiomyoma of the tongue base treated with partial glossectomy performed with harmonic scalpel which shortened surgical procedure, reduced bleeding and postoperative complications.

Case Report

A 76-year old male presented to our department with several months history of globus feeling, without tenderness, dysphagia, dyspnea or dysphonia. Endoscopic ex-

amination revealed a solitary tumor of the right tongue base covered with normal mucosa; sizing 30 by 20 mm. Tumor partially covered the vallecule and dislodged the epiglottis towards endolarynx (Figure 1). Ultrasonographic examination of the neck showed no enlarged lymph nodes. Computed tomography (CT) scan confirmed a

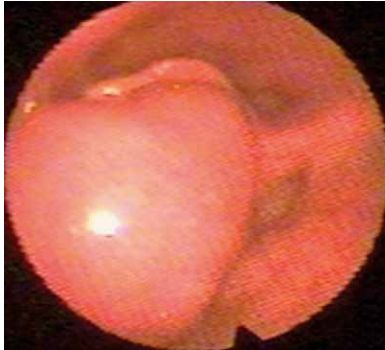


Figure 1. Solitary tumor of the right tongue base with normal mucosa partially covering the vallecule and dislodging the epiglottis towards endolarynx.

well-circumscribed inhomogeneous right tongue base mass which infiltrated glossal muscles. These findings indicated a benign tumor with potentially rich vascularisation. Therefore, diagnostic excision or aspiration biopsy was not performed. With left lateral pharyngotomy approach, we performed a partial resection of the tongue base (Figure 2). Tumor was completely resected with

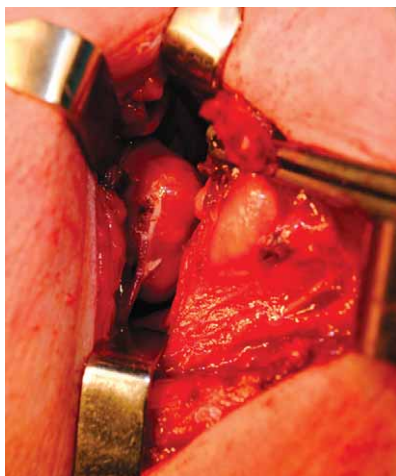


Figure 2. Partial resection of the tongue base with left lateral pharyngotomy approach.

Ultracision Harmonic Scalpel (Eticon Endo-surgery, Cincinnati, USA). There was no perioperative bleeding, and we did not ligate the lingual artery.

Histological examination revealed a well-circumscribed tumor sizing 30x20x15 mm, composed of numerous thick-walled blood vessels and bundles of smooth muscle

cells. These findings were consistent with a diagnosis of angioleiomyoma (Figure 3).

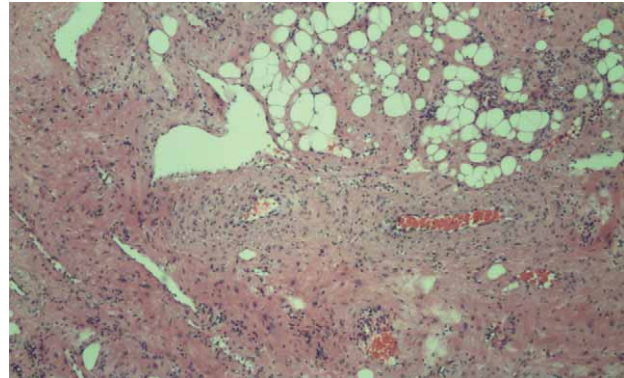


Figure 3. Tumor tissue composed of numerous thick-walled blood vessels and bundles of smooth muscle cells.

Complete epithelialization of tongue lesion occurred in 3 weeks. A 14 months follow-up clinical examination showed no tumor recurrence and patient was completely symptom free.

Discussion

We reported a very rare case of angioleiomyoma of the tongue base. To date there are only 16 cases of angioleiomyoma of the tongue published in the literature (Table 1)^{4,5,7-11}. They usually arise from tunica media of blood vessels but they can also develop out of circumvallated papilla, lingual duct or embryonic muscular tissue. It is generally accepted that angioleiomyoma of the tongue develop from circumvallated papilla's smooth muscle cells⁴. They can be congenital or acquired^{6,12}.

Our patient complained only of globus sensation without any other symptoms. Clinical feature of angioleiomyoma is very nonspecific and these slow growing tumors are usually asymptomatic for a long time. Unlike very

TABLE 1
CURRENT LITERATURE ON ANGIOLEIOMYOMA OF THE TONGUE

Authors	Year	Number of cases
BROOKS JK, NIKITAKIS NG, GOODMAN NJ, LEVY BA	2002	10
MARDEN FA, CALILAO GC, GUZMAN G, ROY SS	2004	1
CHIANG YC, CHEN RM, CHAO PZ, YANG TH, LEE FP	2007	1
GONZALES IP, GUTIERREZ LMJ, LISSET CA, RODRIGEZ JCDV, RENEDO PV	2008	1
KIM YH, JANG YW, PAI H, KIM SG	2010	1
DARLING MR, WEHRLI B, ZELIGMAN E, SMILLIE J, DALEY T	2010	2

painful angioleiomyoma of extremities or trunk angioleiomyoma of the tongue is usually painless. Such unspecific clinical presentation may mimic different other lesions, such as inflammatory disease, benign mesenchymal tumors, soft tissue cystic lesions and vascular lesions^{4,9}.

Owing to its slow growth almost all angioleiomyomas sized less than 20 mm at the time of diagnosis. Therefore, fine needle aspiration biopsy was not required and complete excision of the tumor was both diagnostic and therapeutic procedure. Patohistological and imunohistochemical analysis of the tumor tissue will yield a definite diagnosis. It is very important to distinguish these tumors from their malignant forms, leiomyosarcomas. No case of malignant transformation of oral angioleiomyoma was reported to date.

Despite its good vascularization, no cases of significant perioperative bleeding were reported. Marden et al. reported a hypervascularized tumor of the left side of the tongue sizing 32x30x12 mm that bled significantly (500 ml) during diagnostic biopsy. They performed preoperative diagnostic angiography and selective lingual artery embolisation⁵. Although our patient had one of the largest tumors reported (30 mm) with CT scans indicating vascular nature of the tumor, we did not perform preoperative aspiration biopsy or selective angiography. We decided to perform complete excision with a harmonic scalpel, based on our experience with ultracision technology. In last 5 years we used harmonic scalpel in more than 400 patients with benign or malignant head and neck le-

sions, including 21 patient with tongue tumors. In 2000 Sherman et al. first described partial glossectomy with harmonic scalpel¹¹. To date more than 50 cases with benign or malignant tongue tumors were treated with harmonic scalpel^{13–15}. Analogous to these reports, we also encountered minimal intraoperative bleeding, with no postoperative hematoma or seroma. With harmonic scalpel we resected fractions of tongue tissue up to 5 mm in length, and coagulated blood vessels up to 3 mm in diameter. Thus, we achieved a fast, feasible and almost bloodless surgical procedure. We did not have to perform ligation of the lingual artery or bipolar or monopolar electrocoagulation because of superficial setting of the tumor. Harmonic scalpel induces less destruction of surrounding tissue because of low temperature coaptive coagulation technology (50–100 °C), in contrast to high temperature mono and bipolar electrocoagulation (150–400 °C). In result, there are less necrotic resected margins, wound healing is faster, and material for pathohistological analysis is more preserved. In addition, postoperative pain was lower, shorter, and treated successfully with lower doses of analgesics. Oral feeding was achieved earlier, significantly improving patient's quality of life.

In conclusion, tongue angioleiomyoma are extremely rare, benign, vascular tumors characterized with slow growth and nonspecific clinical presentation. Surgical treatment with harmonic scalpel was proven to be safe and useful in reducing intraoperative and postoperative complications and recovery time.

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KIRURŠKO LIJEČENJE ANGIOLEIOMIOMA JEZIKA ULTRAZVUČNIM NOŽEM – PRIKAZ SLUČAJA I PREGLED LITERATURE

S A Ž E T A K

Angioleiomiomi jezika rijetki su dobroćudni tumori koje karakterizira spori rast, nespecifična simptomatologija, bez zabilježene maligne transformacije. Tumori su građeni od glatkih mišićnih stanica i krvnih žila, a kompletna kirurška resekcija metoda je izbora u terapiji. Parcijalna glosektomija s resekcijom tumora uporabom ultrazvučnog noža pokazala se jednostavnim, brzim i beskrvnim kirurškim postupkom.