Intraoral Excision of Giant Sublingual Angiomyxoma

The Editor,

Sir,

A 29-year-old male patient presented with a painless, slow-growing, solid mass in the left sublingual region, present for 4 months. Clinical examination showed a submucosa mass in the left sublingual region (Figure 1A).

![Figure 1A: Preoperative view of the lesion of the left floor of the mouth. B) Axial contrast-enhanced CT scan demonstrated a well-circumscribed, enhancing solid mass in the left sublingual region.](image1)

The lesion was well demarcated, slightly hard to palpation, and covered by normal mucosa. Ultrasound revealed a well circumscribed, solid mass of approximately 6.5x4.7 cm, located between the left submandibular and sublingual glands. Computed tomography scan showed a well-defined, enhancing solid mass, measuring 6.5x5 cm, and located in the left floor of the mouth. (Figure 1B). Pre-operative informed consent was obtained from the patient. The patient was operated via transoral approach under general anesthesia. The mass was encapsulated and completely removed with submucosal dissection (Figure 2).

![Figure 2A: Intraoperative the view of the mass. B) Postoperatively, the appearance of lobulated, well-circumscribed tumoral specimen.](image2)

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Histopathologic examination of the mass revealed that the tumor composed of relatively uniform, small, stellate and spindled cells, set in myxoedematous matrix with scattered vessels of varying caliber. There was no nuclear atypia or mitotic activity and necrosis. Immunohistochemically, the tumor cells were positive for cluster of differentiation (CD) 34 and negative for S-100 protein and muscle-specific actin (Figure 3). The histopathologic diagnosis was reported as angiomyxoma. Postoperatively, no recurrence was seen during the 12-month follow-up period.

Fig. 3A: Tumour composed of relatively uniform, small, stellate and spindled cells, set in myxoedematous matrix with scattered vessels of varying caliber. (H&E x 100) B) Cytoplasmic reactivity with immunohistochemical CD34 stain in tumor cells. (IHC x 100)

Angiomyxomas are a myxoid mesenchymal tumors that frequently present in the soft tissues of the pelvis and perineal regions of reproductive females. Although there have been many reported cases in the head and neck, only a few angiomyxoma of the oral cavity have been reported in the literature (1–5). Angiomyxomas are most common in middle age, but they may present at any age. There is a slight male predilection (1, 3). The clinical features of angiomyxoma are not pathognomonic and are similar to many of the sublingual and submandibular masses. Ultrasound, computed tomography, and magnetic resonance imaging are helpful to define the lesion in terms of size, location and extension. The definitive diagnosis is performed with histopathologic examination of the lesion. Histologically, the tumor was characterized by spindle-shaped and stellate cells in a loose myxoid stroma, and a
prominent vascular component. Immunohistochemically, angiomyxomas are generally positive for desmin, vimentin, CD34, and α-smooth muscle actin. They are negative for S-100 protein and muscle-specific antigen (1–4). The differential diagnosis should include myxoid neurofibroma, myxoid liposarcoma, myxoid neurofibroma, epidermoid cysts, lipomas, lymphangioma, fibroma, and other soft tissue tumors (1–5). Surgery is the most effective method for the treatment of angiomyxomas. The sublingual angiomyxoma can usually be excised transoral approach and transcervical approach or through combined transoral and transcervical approaches. Angiomyxomas have a high rate of local recurrence between 20% and 40% after primary excision, often due to incomplete excision. Therefore tumor excision with wide tumor-free margins should be performed for these tumors and should be performed long-term follow-up (1–4).

**Keywords:** Angiomyxoma, intraoral excision, recurrence

*H Yaman¹, M Oktay², E Ilhan¹, FH Besir³, E Guclu¹*

*From:¹ Duzce University Duzce Medical Faculty, Department of Otorhinolaryngology  
²Duzce University Duzce Medical Faculty, Department of Pathology ³From Duzce University Duzce Medical Faculty, Department of Radiology*

*Correspondence: Dr H Yaman, Duzce Universitesi Duzce Medical Faculty, Department of Otorhinolaryngology, Duzce/Turkey. Fax: 00903805421387, e-mail: hyaman1975@yahoo.com*
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