

Spontaneous sublingual space hematoma secondary to hypertension: A case report and review of literature

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ABSTRACT

Sublingual hematoma is a rare complication of oral anticoagulant use. Spontaneous sublingual hematoma secondary to hypertension is even rarer and their management remains enigmatic. We present such a case that we successfully managed conservatively along with a review of relevant literatures.

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INTRODUCTION

Spontaneous hematomas in the deep spaces of neck are rare in this age of antibiotics and modern dental care. It has been described as a potential life-threatening complication in anticoagulant users.^[1] Sublingual hematoma, also described as 'Pseudo-Ludwig's' phenomenon, has been described in 1978 by Lepore in patients with coagulation disorders.^[2] It has been described in patients with ill-fitting dentures.^[3] However, to the best of our knowledge, spontaneous sublingual hematoma secondary to hypertension, in the absence of coagulation disorders, being managed conservatively, has not been described yet. Here, we describe a case of spontaneous sublingual hematoma, secondary to hypertension.

CASE REPORT

A 65-year-old male patient presented in the emergency room complaining of a sudden progressive swelling of the floor of mouth with hemorrhagic discoloration. He complained of difficulty in speech and swallowing with mild respiratory distress on exertion. He denied any history of trauma, denture use, anticoagulant use, or tooth infection. He was a known hypertensive and missed to take medicines for last few days.

General examination revealed an uncontrolled blood pressure of 200/100 mmHg in supine position at rest. Pulse rate was 90/min, regular. He was maintaining saturation at room air with respiratory rate of 20/min with SpO₂ 97%.

On local examination, the floor of mouth was grossly swollen and tense, with hemorrhagic discoloration suggestive of recent submucosal bleed. It extended to the ventral tongue surface [Figure 1]. Tongue movement was normal. The swelling was mildly tender on palpation and appeared to continue with the submental space in the neck, with hemorrhagic discoloration of the overlying skin till the suprasternal notch [Figure 2].

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A complete coagulation profile of this patient was done and was found to be normal.

Cardiac evaluation done was unremarkable other than the hypertension.

Contrast-enhanced computed tomography (CT) scan was done, which was suggestive of a non-contrast-enhanced space-occupying lesion in the submental region extending upto the right base of tongue and supraglottis and extension into right parapharyngeal space [Figures 3 and 4]. A few subcentimetric cervical lymph nodes were noted of obscure clinical significance. No vascular abnormality was detected on the CT scan. Doppler study of the neck vessels did not indicate flow disturbances. He was advised magnetic resonance (MR) angiogram but was unable to do so due to financial constraints.

A clinical diagnosis of sublingual hematoma secondary to hypertension was made, and he was managed conservatively. Blood pressure was monitored and antihypertensive medications were started and titrated. However, the hematoma seemed to enlarge with resultant

respiratory distress, and so, an urgent tracheotomy was done to secure the airway.

Post stabilization, the patient underwent direct laryngoscopic examination and biopsy from representative areas. Histopathological examination was negative for any underlying malignancy.

He was put on conservative management with strict control of blood pressure. His hematoma resolved spontaneously over 3 weeks. He was then weaned off tracheotomy and recovered without any complications. Patient was followed up for a month posttreatment and was doing well with no recurrence.

DISCUSSION

Sublingual space is a potential space between the mucosa of floor of mouth and mylohyoid muscle and is a part of suprahyoid group of fascial spaces. The sublingual space communicates with submandibular space along



Figure 1: Hematoma over floor of mouth



Figure 2: Skin discoloration over midline of neck



Figure 3: Contrast-enhanced CT scan showing non-enhancing mass in right parapharyngeal space (axial view). CT = computed tomography

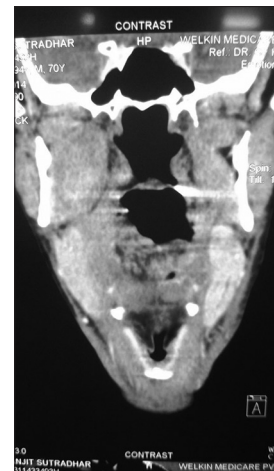


Figure 4: Coronal section showing extension of mass from sublingual space

the posterior border of mylohyoid muscle and below with submental space. Collectively, these three spaces are sometimes termed as perimandibular spaces or submaxillary space. The major vessel of this space is the lingual artery.

Hemorrhage or hematoma of sublingual space can create a “Pseudo- Ludwig’s” phenomenon, in which the tongue and floor of mouth become elevated and can cause airway obstruction.^[2,4] The common causes of sublingual space hematoma are anticoagulant-induced coagulopathy, trauma, or tongue bite.^[1-3,5]

Spontaneous hemorrhage in sublingual space due to uncontrolled hypertension is very unusual and only one such case has been reported to the best of our knowledge.^[6] The etiology of sublingual hematoma in our case is probably the rupture of atherosclerotic vessels supplying the musculature of tongue or other muscles related to the space.

There is no consensus in the management of these patients in current literature. The first step in management is to secure the airway. History taking should include details of anticoagulant use. A careful and thorough study of the coagulation profile of the patient needs to be done. The diagnosis is essentially clinical.

The decision to surgically evacuate sublingual hematoma is controversial with several authors reporting good results even with conservative management once the causative factors have been corrected. They have been found to have spontaneous resolution without surgical intervention once the coagulation profile normalizes.^[5] Use of leeches has also been described to aid in the resolution

of this hematoma.^[7] However, a risk of anaerobic infection is possible, along with the psychological trauma of such intervention. A surgical evacuation would also carry risk of aspiration if the airway has not been previously secured. Some authors believe that surgical attempt to evacuate the blood may cause further swelling and subsequent worsening of condition in postoperative period. We managed our case conservatively, and the patient was successfully treated by 3 weeks of treatment and no complications in the postoperative follow-up.

REFERENCES

1. González-García R, Schoendorff G, Muñoz-Guerra MF, Rodríguez-Campo FJ, Naval-Gías L, Sastre-Pérez J. Upper airway obstruction by sublingual hematoma: A complication of anticoagulation therapy with acenocoumarol. *Am J Otolaryngol* 2006;27:129-32.
2. Lepore M. Upper airway obstruction induced by warfarin sodium. *Arch Otolaryngol Head Neck Surg* 1976;102:505-6.
3. Zeitoun H, Robinson P. A potential hazard for night denture wearers. *J Laryngol Otol* 1994;108:350-1.
4. Hing NR, Bowler MD, Byth PL, Daly CG. Lingual hematoma leading to upper airway obstruction. *Br J Oral Maxillofac Surg* 1985;23:322-5.
5. Pigadas N, Simoes P, Tuffin JR. Massive sublingual haematoma following osseointegrated implant placement in the anterior mandible. *Br Dent J* 2009;206:67-8.
6. Prepageran N, Raman R, Ismail SM, Rahman ZA. Spontaneous sublingual hematoma as a complication of severe hypertension: First report of a case. *Ear Nose Throat J* 2002;81:576-8.
7. Lee NJ, Peckitt NS. Treatment of a sublingual hematoma with medicinal leeches: Report of case. *J Oral Maxillofac Surg* 1996;54:101-3.

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