Rare disease

Spontaneous sublingual haematoma: a rare entity

Mohamed A Bitar,¹ Mohammad A Kamal,² Lorice Mahfoud^{1,3}¹Department of Otolaryngology—Head & Neck Surgery, American University of Beirut Faculty of Medicine & Medical Center, Beirut, Lebanon²American University of Beirut Faculty of Medicine & Medical Center, Beirut, Lebanon³Private practice, Beirut, Lebanon

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Correspondence to Associate Professor Mohamed A Bitar, mb36@aub.edu.lb**Summary**

Sublingual haematoma, also known as pseudo-Ludwig phenomenon, is an entity commonly described in patients on anticoagulation therapy. Spontaneous sublingual haematoma is a rare subtype. It is thought to be due to aneurismal changes in the facial or lingual arteries, occurring mostly in the elderly hypertensive population. Two case reports of spontaneous sublingual haematomas have been reported so far in the literature. Both cases were elderly patients and presented with a sublingual mass and elevation of the floor of the mouth. One patient was intubated whereas the other underwent an emergency tracheotomy. The authors present the third case of spontaneous sublingual haematoma that was afebrile and had a sudden onset of sore throat that progressed to dyspnoea and required a life-saving tracheotomy. The patient recovered quickly postoperatively and could be discharged home in a couple of days and was decannulated in 1 week. No recurrence of the haematoma was noticed on follow-up after 1 year.

BACKGROUND

Sublingual haematoma, also known as pseudo-Ludwig phenomenon, is an entity commonly described in patients on anticoagulation therapy.^{1 2} Spontaneous sublingual haematoma is a rare subtype that occurs in the absence of anticoagulation, with only two cases reported in the literature. Both were elderly patients and presented with a sublingual mass and elevation of the floor of the mouth. One patient was intubated³ whereas the other underwent an emergency tracheotomy.² We present the third case of spontaneous sublingual haematoma that needed a life-saving tracheotomy, to raise awareness about this entity among physicians, especially those who deal with elderly patients who are hypertensive.

CASE PRESENTATION

A patient known to have diabetes mellitus and hypertension presented to the emergency department with a progressive sudden-onset submental swelling over the duration of a few hours resulting in dyspnoea. There was no history of trauma, no recent dental work and no history of anticoagulant intake. There was a history of sore throat prior to the onset of symptoms followed by excessive salivation and slurred speech.

In the emergency room, the patient was awake and oriented but tachypnoeic. Examination revealed a submental tense non-tender swelling in the anterior neck reaching the level of the cricoid and associated with overlying bluish discolouration of the skin. Intraoral examination revealed a sublingual haematoma and elevation of the tongue leading to visual obstruction of the soft palate (figure 1).

INVESTIGATIONS

The patient's blood pressure was 220/100 with oxygen saturation of 95% on supplemental oxygen. Laboratory studies

were within normal range and included complete blood count, chemistry electrolytes panel and coagulation profile.

CT angiography failed to reveal any anomalous vessel or active bleeding (figure 2).

DIFFERENTIAL DIAGNOSIS

- ▶ Trauma
- ▶ Spontaneous bleeding
- ▶ Bleeding due to high blood pressure
- ▶ Bleeding due to anticoagulation intake

TREATMENT

Blood pressure was normalised but the patient's respiratory status continued to deteriorate. A decision was taken to perform controlled awake fibre-optic guided nasotracheal intubation for a pending respiratory failure in the operating

Figure 1 Intraoral examination revealed a sublingual haematoma.

Figure 2 No vascular abnormality or a leak was found.

room. This procedure failed and emergency tracheotomy was contemplated. However, the patient started showing signs of complete airway obstruction with loud inspiratory stridor and oxygen desaturation. A life-saving cricothyroidotomy was performed. After stabilising the patient, the cricothyroidotomy was converted into a regular tracheotomy. After the operation, the patient was transferred to the intensive care unit for observation.

OUTCOME AND FOLLOW-UP

The submental and sublingual swellings decreased at the end of the procedure as blood drained through the incision on the neck. After a couple of days, complete resolution of the haematoma occurred and the larynx could be adequately visualised through flexible fibre-optic laryngoscopy. The patient was discharged home with a tracheotomy tube in place and was decannulated after 1 week. No recurrence of the haematoma was noticed on follow-up after 1 year.

DISCUSSION

Spontaneous sublingual haematoma is quite rare but should be part of the differential diagnosis of sudden-onset oral swelling in elderly patients. It is thought to be due to aneur-

ismal changes in the facial or lingual arteries.² Pre-existing hypertension in our patient probably caused rupture of these weak vessels and resulted in an expanding haematoma. Given the fact that the floor of the mouth is composed of loose soft tissue with multiple potential fascial spaces and is covered only by a layer of mucosa, pooling of blood in these spaces can expand rapidly. This expanding haematoma along with local oedema can displace the tongue posteriorly and lead to airway obstruction. Sore throat appears to be the first sign in such cases,^{1 4} probably because of the expansion of the submucosal area which causes stretching and irritation of the tissues and thus pain.

Rapid intervention is crucial to avoid airway collapse and respiratory arrest. Performing an emergency tracheotomy can be hard and hazardous because the procedure has to be done under local anaesthesia while the patient is awake and often agitated. The haematoma can further dissect into the fascial planes of the lower neck, thus effacing the anatomy of the anterior neck with loss of landmarks during dissection. Moreover, the haematoma can drain profusely during the surgical procedure and obstruct the surgical field, making the procedure more difficult.

Learning points

- ▶ Spontaneous sublingual haematoma is a rare entity.
- ▶ It should be suspected when sudden sore throat occurs in an afebrile hypertensive elderly patient.
- ▶ Rapid diagnosis and management of the airway are imperative to save the patient.

Competing interests None.

Patient consent Not obtained.

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