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## Spontaneous submental hematoma, a pseudo-Ludwig's phenomenon in 101-year-old patient: case report and literature review

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## ABSTRACT

**OBJECTIVE:** Submental hematoma or pseudo-Ludwig's phenomenon, is a rare entity seen in anticoagulated patients and can precipitate upper airway obstruction. Our objective is to present a rare case of spontaneous submental hematoma due to poorly controlled hypertension in elderly patient and to perform a literature review.

**CASE REPORT:** 101-year-old female presented to emergency room with sudden painful swelling in the floor of mouth and slurred speech. Not on anticoagulation and no history of trauma or known allergies. Physical examination and flexible laryngoscope revealed normal temperature and blood pressure of 190/100, submental/floor of mouth swelling that was tense to palpation, ecchymotic/hemorrhagic and extend to the tip of the tongue suggestive of recent submucosal bleeding and mild swelling at the base of tongue as well as small hemorrhagic vallecular cyst. CT scan ruled out AVM and pseudoaneurysm of lingual artery. She was diagnosed with spontaneous submental hematoma (SSH) probably due to the rupture of atherosclerotic vessels supplying the musculature related to the space due to uncontrolled severe hypertension. She was treated conservatively by electively securing the airway and the swelling resolved in 3 days. She got extubated and subsequently discharged home.

**METHODS:** Systematic literature review was conducted and revealed only 5 cases since 2002 reported with this rare subgroup of spontaneous submental hematoma not related to anticoagulation. This case report has been reported in line with the SCARE criteria [1].

**RESULTS:** 4 cases were results of uncontrolled hypertension whereas one case was seen in cirrhotic liver patient. One patient was managed with surgical evacuation of the hematoma were in the others conservative management initiated but failed. Of those 4, 2 required an emergent airway procedure.

**CONCLUSION:** Spontaneous submental hematoma (SSH) is a clinical diagnosis and conservative management is successful in reducing the amount of swelling once the causative factors have been corrected/controlled. The main goal is to electively secure the airway and as in all emergent airway management the team must include members capable of a surgical airway.

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## 1. Background

Sublingual hematoma or pseudo-Ludwig's phenomenon, is a rare entity seen in anticoagulated patients and can precipitate upper airway obstruction. A rare subgroup includes a spontaneous sublingual hematoma felt to be related to poorly controlled hypertension in elderly patients. It thought to be secondary to aneurysmal changes in lingual or facial arteries in this age group. To date there are only 6 cases reported of spontaneous submental hematoma with varying management plans, including: conservative management with close observation in IMCU/ICU,

prophylactically securing the airways with elective intubation and/or emergency tracheostomy. Some authors proposed an evacuation the hematoma through an incision along the floor of the mouth. This case presents the oldest patient reported with SSH a 101-year-old female who recovered uneventfully after conservative management with elective prophylactic fiberoptic intubation. We present this rare entity to raise awareness about this entity among physicians, especially those who deal with hypertensive elderly patients.

## 2. Case report

101 year old female with past medical history of dyslipidemia and hypertension and taking aspirin 81 mg for primary prevention, presented to a regional emergency department complaining of sud-

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**Fig. 1.** Oral examination showing significant floor of mouth with swelling and hemorrhagic changes.

den swelling in the floor of mouth with pain and slurred speech. She has no known allergies and there was no history of trauma. She denied shortness of breath and there was no stridor. She received a dose of dexamethasone 20 mg IV and Benadryl 50 mg IV to reduce the swelling with no improvement. She was subsequently transferred to the QEII HSC, a tertiary hospital in Halifax, NS.

Vital signs were normal on arrival with the exception of a blood pressure of 190/100. Oral cavity exam demonstrated submental/floor of mouth swelling that was tense to palpation, ecchymotic/hemorrhagic and extend to the tip of the tongue suggestive of recent submucosal bleeding. While base of tongue was normal to palpation, palpation of the tip was painful.

The patient was edentulous; gums and buccal mucosa were not involved.

Neck examination was significant for swelling in the submental area and mild discoloration of the skin down to level of supra-sternal notch.

Flexible pharyngolaryngoscopy was performed with evidence of mild swelling at the base of tongue as well as small hemorrhagic vallecular cyst. Supraglottic structures were unremarkable and there was no significant vocal folds edema.

Complete blood count and coagulation profile was found to be normal. Creatinine was slightly elevated to 117 (49–90  $\mu\text{mol/L}$ ) (Fig. 1).

Unenhanced and enhanced CT scan of the neck demonstrates large heterogeneous nonenhancing mass at the floor of the mouth confined within the sublingual space (straight arrows in Fig. 2) which measures approximately  $3.3 \times 3 \times 2.6$  cm in three dimensions. The lesion obliterates the adjacent fat planes between the genioglossus and geniohoid muscles bilaterally. No convincing extension to the adjacent submental or submandibular space beyond the visualized mylohyoid muscles bilaterally (arrowhead in Fig. 2C). It demonstrates significant adjacent superior mass effect on the adjacent tongue (curved arrow in Fig. 2), and the endotracheal tube superolaterally (arrowhead in Fig. 2B and C).

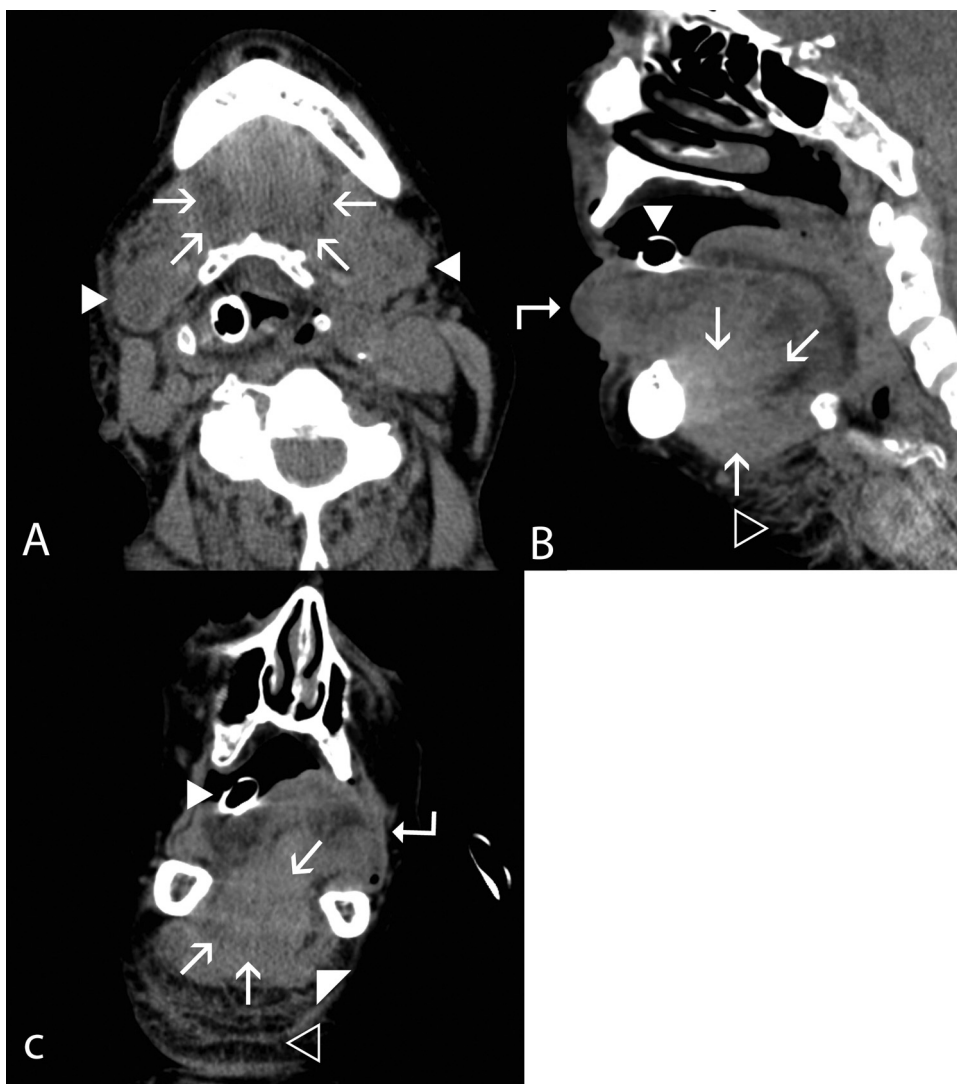
There is a significant adjacent inflammatory fat stranding within the submental space subcutaneous fat inferiorly (open arrowhead in Fig. 2B and C), with submandibular glands heterogeneous enlargement posterolaterally within the submandibular space (arrowhead in Fig. 2A). There are no suspicious enlarged cervical

lymph nodes by CT size criteria to suggest underlying metastasis of a neoplastic process. The remainder of the soft tissues neck is unremarkable. The CTA images demonstrate normal morphology and opacification of lingual arteries bilaterally, with no evidence of abnormal tangle of vessels or arterial pseudoaneurysm to suggest underlying vascular pathology (Fig. 3).

Angioedema was ruled out owing to the skin and mucosal ecchymotic discoloration. Her findings were thought to be consistent with some disturbance or alteration in vascular flow such as an arteriovenous malformation (AVM), pseudoaneurysm, or cavernous hemangioma. These were ruled out with CT angiogram of neck vessels as above. A clinical diagnosis of spontaneous submental hematoma secondary to uncontrolled hypertension was made. Her blood pressure was monitored and antihypertensive medications were started and titrated. She was immediately given high-dose intravenous dexamethasone and nebulized epinephrine to reduce his associated upper airway edema. She was taken to the operating theatre for an awake intubation. In the OR the anesthesia team were able to perform an awake fiberoptic orotracheal intubation. The otolaryngology team was available for an emergent tracheotomy procedure. The patient was subsequently taken to the ICU for post-intubation care and she remained there for 3 days. Once the swelling had subsided and an air leak around the endotracheal tube was obtained she was successfully extubated. Strict blood pressure control was achieved then she was transferred to the floor to optimize her discharge planning. She was discharged 8 days after her presentation. She was followed up 6 weeks after discharge with complete resolution of the swelling and ecchymosis (Fig. 4).

### 3. Discussion

The 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 the posterior border of mylohyoid muscle and below with submental space. Collectively, these three spaces are sometimes termed as peri-mandibular spaces or submaxillary space. The major vessel of this space is the lingual artery. The floor of the mouth is composed of loose soft tissue with



**Fig. 2.** (A) Axial image through the hyperdense mass within the floor of mouth confined to the sublingual space “arrows”, associated with heterogeneous submandibular gland enlargement posterolaterally within the submandibular space “arrowhead”. (B, C) Sagittal and coronal images through the lesion “arrows” demonstrates significant mass effect on the adjacent tongue “curved arrows” and endotracheal tube “arrowhead”, associated with moderate reactive subcutaneous inflammatory fat stranding in the adjacent submental space “open arrowhead”. Note the mylohyoid muscles “curved arrowhead in C” separating the lesion from the submental space inferiorly and the submandibular space posterolaterally.

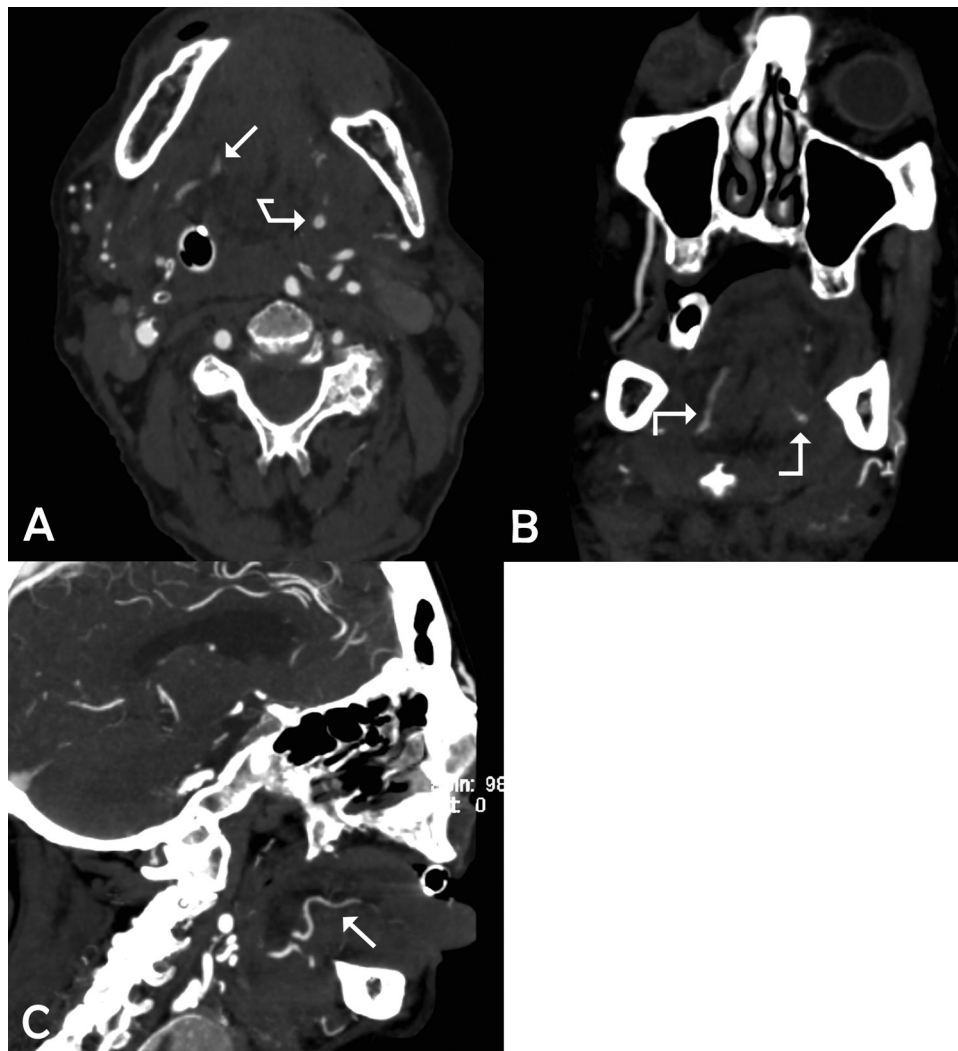
multiple potential fascial spaces and is covered only by a layer of mucosa. Pooling of blood in these spaces can expand rapidly. This expanding hematoma along with local edema can displace the tongue posteriorly and lead to airway obstruction.

Hemorrhage or hematoma of sublingual space can create a “Pseudo-Ludwig’s” phenomenon, which was first described by Lepore in 1976 as, a condition caused by deranged coagulation with warfarin therapy [1]. The floor of the mouth, though highly vascular, is rarely a site of hematoma formation. Floor of mouth hematomas have been described after trauma and dental implant procedures, anticoagulant-induced coagulopathy or uncontrolled hypertension [2–4]. Spontaneous hemorrhage in sublingual space due to uncontrolled hypertension is very unusual and only a few cases have been reported [5–10].

The differential diagnosis for this patient included trauma, hematoma secondary to coagulation abnormalities, AVM, pseudoaneurysm, or angioedema. Trauma seemed highly unlikely, because the patient maintained a soft food diet and sedentary lifestyle. The swelling started bilaterally and briskly, which is an unexpected location for trauma. The patient was edentulous with complete

dentures. The dentures were examined as a possible source of trauma and found to be free from abnormality. Also, there were no intraoral lacerations or abrasions.

Pseudoaneurysm forms as the result of separation in between the tunica intima and adventitia in an artery. Pseudoaneurysm and arteriovenous malformation (AVM) were excluded based on CT neck angiogram study [11]. Angioedema is a rapid swelling of the dermal, subcutaneous and submucosal tissues often accompanied with urticaria, pruritus, and pain, usually caused by an allergen, angiotensin-converting enzyme (ACE) inhibitor, or angiotensin receptor blocker. Treatment of choice is usually antihistamines, steroids, or epinephrine. Based on the finding in our patient this was excluded as she was on hydrochlorothiazide for her blood pressure control and her examination showed hemorrhagic/ecchymotic mucosal appearance and no urticaria was found on skin exam. Genial tubercle fracture can be another causative agent and should be included in the differential diagnosis for spontaneous bilateral sublingual hematoma, as reported in 1894 [7,13]. Typically, if a genial tubercle fracture is suspected, CT is used to confirm the diag-



**Fig. 3.** (A) CTA axial images through the lesion demonstrate no convincing active extravasation of the IV contrast from the lingual arteries bilaterally “arrows”. The rest of the visualized external carotid artery branches are within the normal limits as well. (B, C) Coronal and sagittal images through the lingual arteries “arrows” didn’t show any underlying abnormal tangle of vessels or arterial pseudoaneurysm to suggest underlying vascular pathology.

nosis. Genital tubercle fracture was not noted on CT scan of our patient.

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 due to uncontrolled severe hypertension. Proper history and physical examination is fundamental and careful inquiry about anti-coagulation use is crucial. Clinicians must study the coagulation profile and manage it accordingly. The diagnosis is essentially clinical [3–5].

There is no consensus among previous reports in the management of these patients. The first step in management is to secure the airway. Conservative management was successful in reducing the amount of swelling once the causative factors have been corrected/controlled. Even in coagulopathy-induced hematoma, there has been spontaneous resolution without surgical intervention once the coagulation profile normalizes [3]. The decision to surgically evacuate sublingual hematomas is controversial. Some authors believe that surgical attempt to evacuate the blood may cause further swelling and subsequent worsening of condition in postoperative period [6,8]. Use of leeches has also been described to aid in the resolution of this hematoma. However, the risk of anaerobic infection and psychological burden on the patient must be considered [12].

We managed our case conservatively, and the patient was successfully treated with strict blood pressure control and electively securing the airway while the swelling continued to worsen initially. It is crucial to act proactive to protect the airways when feasible and decide if the surgical intervention required thereafter. And as in all emergent airway management the team must include members capable of a surgical airway.

#### Competing interests

None.

#### Funding

None.

#### Ethics approval

Not applicable.



**Fig. 4.** Ongoing floor of mouth swelling after intubation. Notice the skin changes with ecchymosis and bruises that lags few days from the onset of the symptoms (A, B). Marked improvement and reduction in the swelling after few days of intubation, skin bruises worsen before start improving (C).

### Consent

Patient provided informed consent for publication of the case report. Editor-in-chief will be provided with a copy of the consent upon request at any time.

### Author contributions

Uthman Alamoudi and Yasser Alsallumi: collected data, wrote article; Robert Hart: designed study, revised article; Mathew Rigby: analyzed data; S. Mark Taylor: designed study, revised article; Jonathan Trites: designed study, revised article.

### Guarantor

Uthman Alamoudi.  
Robert D. Hart.

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