



Case Report

Traumatic lingual haematoma: Another unusual cause of upper airway obstruction in systemic lupus erythematosus

M^aFe García Reija^{a,*}, Marcos Fernández-Barriales^b, Tomás González Terán^a, Sergio Sánchez Santolino^a

^a Servicio de Cirugía Oral y Maxilofacial, Hospital Universitario Marqués de Valdecilla, Avenida de Valdecilla S/N. 39008 Santander, Spain

^b Servicio de Cirugía Oral y Maxilofacial, Hospital Universitario de Araba, 01006 Vitoria-Gasteiz, Spain

ARTICLE INFO

Keywords:

Chronic renal failure
Lingual haematoma
Systemic lupus erythematosus
Oral hemorrhage

ABSTRACT

Lingual hematoma (LH) is an uncommon and potentially life-threatening condition due to its tendency to cause upper airway obstruction. It usually occurs as a result of trauma (motor vehicle accidents, grand mal seizures or traumatic tracheal intubations) and rarely spontaneously in cases of patients with inherited or acquired coagulopathies, high blood pressure, hematological disorders, or vascular malformations.

Herein, we report the first case, to our knowledge, of a traumatic massive lingual hematoma in a patient with Systemic lupus erythematosus (SLE) secondary to tongue biting after neurological deterioration, hypertensive crisis and multiple tonic clonic seizures during hemodialysis for chronic renal failure.

Introduction

Systemic lupus erythematosus (SLE) is an inflammatory autoimmune disorder that affects multiple organ systems with main involvements in kidney, skin and blood. It is usually present in young adult women and is diagnosed by the presence of standard criteria.

Symptoms and signs depend on which organs are involved. Hemorrhages are a dangerous and potentially life-threatening complication in SLE. The most common etiologies are hypertension, anticoagulation, catastrophic anti phospholipid antibody syndrome (APLA) and comorbidities.

Alveolar pulmonar hemorrhage, spinal hemorrhage, intracranial hematomas, gastrointestinal hemorrhage, retroperitoneal, inner ear or even acute hemorrhagic myocarditis [1–5] has been previously reported in the literature in patients with SLE, however, to the best of our knowledge, there are no published case reports of massive lingual hemorrhage.

In this article, we will describe the etiology, diagnosis and management of upper airway obstruction due to traumatic lingual hematoma in a patient with SLE. This is an interesting case of a patient with some of the numerous complications associated with chronic renal failure which in combination resulted in an unusual cause of life-threatening upper airway obstruction.

Case report

A 27-year-old man was admitted to the Emergency Department after acute neurological deterioration, tonic-clonic seizures and

* Corresponding author at: Calle Ramón y Cajal 43-4^ªsDerecha, 39005 Santander, Spain.

E-mail address: mfe.garcia@scsalud.es (M.G. Reija).



Image 1. Massive swelling of the tongue protruding out of his mouth. Lateral view.

hypertensive crisis during hemodialysis.

His past medical history was significant for a seven year history of SLE with hypertension and a long standing lupus-related Chronic Kidney Disease in renal replacement therapy of 3 years' duration after acute rejection of kidney transplant. He was on treatment with amlodipin 5 mg, doxazosin 8 mg, carvedilol 25 mg, enalapril 25 mg and omeprazole 20 mg with poor adherence to medication.

On examination, he was afebrile, responsive to verbal stimuli and oriented in time, place, and person with no evidence of apparent focal deficits. His blood pressure was 200/120 mmHg, pulse 132/min and the pulse oximeter was reading at 98%. Cardiovascular and respiratory system examination revealed no abnormality. The electrocardiogram was normal. Laboratory tests on presentation were remarkable for elevated urea and creatinine of 116 mg/dl and 12,61 mg/dl respectively and thrombocytopenia with a platelet count of 131×10^3 per μL . The coagulation profile was unremarkable with a PT of 100 s and a APTT of 28 s.

One hour later, the patient was conscious and breathing comfortably, when he suffered a generalized convulsion. Immediate management included intravenous benzodiazepines (diazepam 5 mg) followed by 1000 mg Levetiracetam without response, so he received the administration of further diazepam dosage. He was admitted to the Intensive Care Unit (ICU) for more aggressive management under assisted ventilation. Patient presented at the ICU with severe dyspnoea and massive swelling of the tongue. The patient was finally anaesthetized with propofol and fentanyl infusions and intubated to secure airway.

After airway management was achieved by orotracheal intubation, physical examination revealed a massive swelling of the tongue which was significantly displaced anteriorly and superiorly, protruding out of his mouth 3 cm (Images 1,2), firm on palpation with a generalized dark-blue coloration. The dorsal surface of the tongue showed a 1,5 cm laceration on the posterior right half which was sutured. No hemorrhage was observed other than that of lingual hematoma.

A computed tomography scan of the head and neck showed severe diffuse enlargement of the tongue with obliteration of the airway without any particular vessel to be the cause of the hematoma. Thus embolization was not attempted because the source of bleeding was not identified. It was thought that the vessels had undergone tamponade from physical pressure from the hematoma and thus there was likely no active hemorrhage.

A clinical diagnosis of lingual hematoma secondary to trauma (tongue biting), hypertension and thrombocytopenia was made, and he was managed conservatively with intravenous midazolam and levetiracetam to prevent further seizure activity. Blood pressure was monitored and antihypertensive medications were started and titrated. Percutaneous tracheotomy was performed to secure the airway,

On the third day of admission the patient was diagnosed of Ventilator-Associated Pneumonia caused by *Enterobacter aerogenes*. According to the antibiotic susceptibility pattern of the isolated bacteria he was treated with meropenem (2 g every 8 h) for 10 days. The patient's lingual hematoma resolved over five to six days. The blood pressure was well controlled near 140/90 mmHg with a combination of several anti-hypertensive medication. The patient was discharged on hospital day 34th. Follow-up at 2 weeks showed complete resolution of the lingual hematoma and traumatic ulcer (Image 3).

Discussion

Lingual hematoma is both a rare and potential life-threatening phenomenon due to its tendency to cause upper airway obstruction. Bleeding from the lingual artery or its branches can result in very drastic tongue enlargement. This enlargement results in the tongue being displaced in a cephalad and posterior direction endangering the patient's life. A classification of acute enlargement of the tongue has been formulated by Renehan and Morton [6] and is based on the various etiologies encountered. Their classification



Image 2. Lingual haematoma. Frontal view.



Image 3. Complete resolution of the lingual hematoma and traumatic ulcer.

system includes hemorrhage secondary to trauma, vascular anomaly or disorder of coagulation, edema secondary to exudates or transudates, infarction, and infection. In this case, the patient had obviously suffered a lingual hematoma secondary to two subclasses of etiology (trauma and coagulopathy), both mediated by chronic renal failure, hypertensive crisis and multiple tonic clonic seizures in association with Systemic Lupus.

Airway management is of prime importance in cases of acute tongue enlargement. Progressive lingual and sublingual swelling displaces the tongue posteriorly and cephalad eventually producing dysphonia, drooling, dyspnoea and finally stridor heralding upper airway obstruction. In the presence of these features, it is axiomatic that a definitive airway must be established. Endotracheal intubation is often difficult to perform orally in such cases and thus is usually performed nasally. Blind nasal intubation can be extremely difficult and potentially traumatic. Fiberoptic laryngoscopy can be very helpful except in cases of active hemorrhage obstructing the view. Given the difficulty of such intubations, they are often performed with the patient awake because the ability to ventilate a patient with a bag-valve mask is unpredictable. Depending on the surgeon's experience, a cricothyroidotomy or a rapid emergent tracheostomy can be used for rapid airway establishment [7]. In our patient the decision was made to perform an immediate endotracheal oral intubation with the patient awake. Oral intubation was successfully completed on the first attempt.

After airway management is achieved, the most critical step management is recognition of the event itself. Therefore, a careful

evaluation of clinical signs and laboratory data is essential in order to make the right medical decision. In cases with diffuse hemorrhage, a hematological disorder should be kept in mind. Those presenting with active SLE can present with immune thrombocytopenia. In our particular case of a patient with some of the numerous complications associated with SLE and chronic renal failure, the hemorrhage was due not only to decreased platelet count, but also to platelet dysfunction as a result of intrinsic platelet abnormalities and impaired platelet-vessel wall interaction [8]. This probably led to the unusually large haematoma of the tongue when he bit it, sufficient to cause an acute upper airway obstruction. The profuse bleeding associated with high blood pressure also contributed to the airway emergency.

The management of these cases can vary depending on the cause of the hemorrhage. In cases with active bleeding hemostasis can be obtained by local control, interventional radiology or ligation of injured vessels. Hematoma evacuation of the tongue is not usually indicated because bleeding occurs into the intrinsic muscles of the tongue rather than into the potential anatomic fascial spaces [8]. Some authors believe that surgical attempt to evacuate the blood may cause further swelling and subsequent worsening of condition in postoperative period. Those due to inherited or acquired coagulopathies, treatment is usually conservative once the causative factors have been corrected. However, there is some controversy in cases in which patients undergo anticoagulation therapy acutely. Some of these cases are managed with anticoagulation reversal, whereas other authors prefer observation while maintaining anticoagulation to prevent rethrombosis of the targeted vessels [9].

In conclusion, lingual hematomas can be a deadly phenomenon requiring rapid identification and management. The first objective of treatment should be guarantee airway safety. Once the airway is secured the treatment focus switches to hemostasis and etiology assessment. In our particular case, decreased platelet count and probable platelet dysfunction associated with trauma and uncontrolled hypertension, played a significant role in the development of the lingual hematoma.

References

- [1] M.E. Cucuzza, S.D. Marino, L. Schiavone, P. Smilari, F. Filasco, P. Barone, Diffuse alveolar haemorrhage as initial presentation of systemic lupus erythematosus: a case report, *Lupus* 27 (3) (2018) 507–510.
- [2] M.C. Abdulla, J. Alungal, S. Hashim, M.M. Ali, M. Musambil, SLE presenting as multiple hemorrhagic complications, *Lupus* 24 (10) (2015) 1103–1106, <https://doi.org/10.1177/0961203315573853> (Epub 2015 Feb 24).
- [3] K.G. Goh, S.G. Ong, Recurrent spontaneous subdural hematoma secondary to immune thrombocytopenia in a patient with overlap syndrome, *Lupus* 24 (1) (2015) 90–93, <https://doi.org/10.1177/0961203314554248> (Epub 2014 Oct 10).
- [4] P. Dickens, J. Nicholls, C.P. Lau, Acute hemorrhagic myocarditis in systemic lupus erythematosus, *Heart Vessel*. 7 (2) (1992) 104–106.
- [5] M. Sugiura, S. Naganawa, M. Teranishi, E. Sato, S. Kojima, T. Nakashima, Inner ear hemorrhage in systemic lupus erythematosus, *Laryngoscope* 116 (5) (2006) 826–828.
- [6] A. Renehan, M. Morton, Acute enlargement of the tongue, *Br. J. Oral Maxillofac. Surg.* 31 (1993) 321–324.
- [7] H.S. Dhaliwal, S.S. Dhaliwal, R.D. Heckel, F.A. Queresby, D.A. Baur, Diagnosis and management of upper airway obstruction due to lingual hematoma: report of a case, *J. Oral Maxillofac. Surg.* 69 (2) (2011) 558–563.
- [8] K.P. Ng, Lingual haematoma: yet another unusual cause of upper airway obstruction, *Med J Malaysia* 53 (1) (1998) 112–114 Mar.
- [9] Z. Song, B. Laggan, A. Parulis, Lingual hematoma treatment rationales: a case report, *J. Oral Maxillofac. Surg.* 66 (3) (2008) 535–539.