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TRAUMATIC LINGUAL HEMATOMA RESULTING IN BILATERAL TEMPORAL MANDIBULAR JOINT DISLOCATIONS

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☐ Abstract—Background: Lingual hematoma (LH) is a relatively uncommon entity seen after both medical and traumatic etiologies. Regardless of the cause, the feared complication is acute airway obstruction. Case Report: Our case involves a 39-year-old man who presented to the Emergency Department via emergency medical services with an enlarging LH after an unwitnessed fall, suspected to be an alcohol withdrawal seizure. The bleeding was likely exacerbated by previously undiagnosed thrombocytopenia. Airway stabilization was rapidly established via nasotracheal intubation after standard intubation techniques were deemed unfeasible. Despite correction of the coagulopathy, the LH continued to expand, resulting in bilateral tympanomandibular joint (TMJ) dislocations. To our knowledge, this complication has not been previously reported as a complication of LH. Why Should an Emergency Physician Be Aware of This?: Despite being a relatively uncommon condition, LH has the potential to result in life-threatening airway obstruction with limited airway options. Prompt airway stabilization should be the first priority upon diagnosis. A rapidly evolving LH can limit standard orotracheal rapid sequence intubation options, and may require alternative airway procedures. Additionally, ongoing lingual swelling after airway stabilization has now been shown in our case to result in bilateral TMJ dislocations. Concurrent management of reversible coagulopathy may help prevent this complication or reduce its severity. © 2018 Elsevier Inc. All rights reserved.

☐ Keywords—lingual hematoma; airway obstruction; TMJ dislocation; coagulopathy; nasotracheal intubation

INTRODUCTION

Traumatic lingual hematoma (LH) is an uncommon condition that can rapidly progress to life-threatening upper-airway obstruction. The tongue is a richly vascular structure, mainly supplied by branches of the lingual artery. The majority of LHs are traumatic or spontaneous; the latter are often associated with the use of thrombolytics or other anticoagulants. To our knowledge, there are no reports documenting bilateral TMJ dislocation as a complication. We report a case of traumatic LH complicated by thrombocytopenia, which necessitated airway management with limited options. We will discuss airway stabilization in the setting of a large anterior airway obstruction, treatment of a traumatic LH, and a previously unreported complication of LH resulting in bilateral TMJ dislocations.

CASE REPORT

A 39-year-old man with a history of alcohol abuse was brought into the Emergency Department (ED) by

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ambulance due to an unwitnessed fall and suspected alcohol withdrawal seizure. Upon arrival the patient was alert and oriented with no evidence of acute respiratory compromise, despite a muffled voice and a large LH. He had no complaints and denied shortness of breath. Past medical history was significant only for alcohol abuse, but he had not imbibed for 24 h. He denied use anticoagulants, angiotensin-converting enzyme (ACE) inhibitors, or history of allergic reaction. He also denied a prior history of tongue swelling or difficulty closing his mouth. He stated that both started while he was being transported to the hospital and were progressively getting worse. On arrival, the patient had a heart rate of 155 beats/min, temperature of 37.9°C, respiratory rate of 17 breaths/min, room air oxygen saturation of 100%, and a blood pressure of 191/121 mm Hg.

Examination revealed a tremulous man with a circumferential dressing covering a small forehead laceration. He appeared to be withdrawing from alcohol, but was otherwise surprisingly comfortable appearing. He was unable to completely close his mouth due to a very large, ecchymotic tongue that occupied his entire oral cavity, but he was able to reduce his maximum incisor opening about ½ cm when asked. His secretions were well controlled and he spoke in full sentences without stridor. It was impossible to visualize his posterior pharynx or fully insert a tongue depressor due to the LH size. Scant bloody intraoral secretions were noted as the tongue became traumatized from superficial lacerations as it continued to expand into the molars. Palpation of his TMJs did not reveal significant subluxation or dislocation. He preferred to sit upright, but was not tripoding. Lungs were clear to auscultation bilaterally. It was apparent that this patient was at risk for airway compromise, and intubation was initiated.

This patient's presentation, coupled with his vital signs, triggered activation of the trauma service. Thus, there was a team of both emergency physicians and trauma surgeons at the bedside. As soon as we began discussing airway management, the trauma team asked us to not intubate the patient, as "he appeared stable." It was pointed out to the trauma team that there was a potential of the LH expanding to the point where it might dislocate his TMJs, and regardless, airway stabilization was mandatory. This conversation prompted a careful TMJ examination and at that time, there was no suggestion of TMJ subluxation or dislocation.

Initially, 2 mg of lorazepam i.v. was administered for seizure prophylaxis and to abate the hyperadrenergic state. This was followed by the administration of 1 mg/kg of ketamine i.v. Paralytics were contraindicated given that no device could be inserted into the oral cavity past the LH, and bag-valve-mask ventilation was anticipated to be unsuccessful. Glidescope and direct laryngoscopy intubation

were not possible, as the expanding LH prevented their insertion. Fiber-optic scope was not immediately available in the ED, and a surgical airway was deemed not optimal unless a more conservative approach failed. The decision was made to attempt nasotracheal intubation with the patient seated and mildly sedated. A 5.5-cm endotracheal tube was introduced into the trachea via the nares. Placement was confirmed with auscultation, capnography, oxygen saturation, and chest radiography. The patient was then heavily sedated with a midazolam and fentanyl drip. Oxygen saturation remained 100% throughout the procedure. Laboratory results post intubation revealed a prothrombin time/international normalized ratio of 15.7/ 1.3, respectively, and a platelet count of 28,000. A postintubation maxillofacial computed tomography scan revealed bilateral TMJ subluxations, which eventually progressed to bilateral dislocations.

The patient was admitted to the medical intensive care unit for medical management. Multiple attempts to reduce the TMJ dislocations by head and neck surgery as well as oral-maxillofacial consultants over the ensuing week were unsuccessful until hospital day 7. No surgical procedures were required. He was extubated on day 14. His course required long-term, deep sedation for protracted seizures, likely related to alcohol withdrawal. Consultants believed that the seizures continued to induce lingual trauma and impede its resolution. An electroencephalogram showed diffuse slowing without other abnormalities, and magnetic resonance imaging of the brain was normal. He was discharged on hospital day 18, neurologically intact except for generalized weakness related to long-term intubation, necessitating a walker.

DISCUSSION

Traumatic LH is a rare condition, with airway obstruction being the feared complication. Resolution of glossomegaly can take days. Most reports of LH result from thrombolytics, anticoagulants, or hemophilia, and are of a spontaneous nature. Fewer reports discuss LH resulting from trauma (1). Traumatic cases have been associated with seizures and other types of oral-facial trauma or instrumentation such as dental procedures. To our knowledge, there are no reported cases of LH resulting in bilateral TMJ dislocation.

The tongue is an extremely vascular structure comprised of both intrinsic and extrinsic muscles and supplied by branches of the lingual artery, off the external carotid artery. LH resulting in severe airway compromise is rare, but airway management should always be first priority. If the LH rapidly enlarges, it can impede the airway as it continues to displace the tongue both superiorly and posteriorly. Less commonly, bleeding can originate in or spread to the sublingual and submandibular spaces.

Lepore likened this to Ludwig's angina and termed it "pseudo-Ludwig" phenomenon (2).

The presence of symptoms such as dysphagia, drooling, or stridor indicate the need for an artificial airway. In the presence of these features, it is axiomatic that an early definitive airway be established. Preferred airway techniques such as rapid sequence oral intubation can be difficult or impossible, as noted in this case. Further, paralyzing a patient with a large, obstructive tongue can result in difficulty oxygenating. Fiber-optic laryngoscopy through the nares can be very helpful, except in cases of active hemorrhage, but this equipment may not be readily available in the ED. Sedation without the use of paralytics is useful when visualization of the oropharynx is felt to be possible. Ketamine was administered to this patient, as it does not suppress the airway's protective reflexes and allows for continued spontaneous respiration. It was preceded by benzodiazepine administration for seizure prophylaxis and sedation.

In recent decades, nasotracheal intubation has fallen out of favor, but this case demonstrates that it can still have a role when oral approaches are not feasible (3). Blind nasotracheal intubation can be traumatic as the tube passes through the nose, and known coagulopathy is considered a contraindication, however, in this case, the coagulopathy was unknown initially. Intentionally selecting a smaller (5.5 cm) endotracheal tube allowed for a less traumatic insertion as opposed to a larger tube. The tube can later be changed to a larger size, if desired, over a fiber-optic scope. Nasotracheal intubations are often performed with the patient awake or mildly sedated rather than paralyzed, because the ability to ventilate a paralyzed patient with an airway obstruction using a bag-valve mask is unpredictable. Additionally, nasotracheal intubation is ideally done in a sitting patient, which is often less emotionally traumatic for them. When conservative airways are not possible, a surgical airway may be indicated. The possibility of an existing coagulopathy should be considered during any surgical procedure. Cricothyroidotomy is a surgical option for establishing the airway in a patient with an expanding LH, however, it requires the patient to be adequately sedated to allow a supine position for the procedure. Tracheostomy is so rarely performed in the ED and is inherently a lengthy and more difficult procedure, that it is unlikely to be a necessary or viable option. Jet insufflation is not indicated with a large, upper-airway obstruction, as inadequate exhalation can result in barotrauma.

TMJ dislocation can result from trauma or non-traumatic situations. To date, it has not been a reported complication after either type of LH. If unable to reduce the dislocation acutely, it may be prudent to wait for some resolution of the glossomegaly. Interestingly, this is the second time one author (KN) has seen a bilateral TMJ dislocation resulting from glossomegaly. The first non-traumatic case was related to ACE inhibitor angioedema, leading us to believe that perhaps this complication is underappreciated or underreported.

A search for a contributing coagulopathy should be undertaken and reversed when identified. In the rare situation, ipsilateral, extraoral ligation of the lingual artery may be required for active, life-threatening lingual bleeding once the airway has been secured. This would necessitate emergent surgical intervention. Lingual bleeding can continue after ligation due to extensive collateral circulation (1). Steroids play a role in the treatment of glossomegaly resulting from anaphylaxis or angioedema, but there are no studies showing efficacy in their use to treat traumatic LH.

WHY SHOULD AN EMERGENCY PHYSICIAN BE AWARE OF THIS?

In conclusion, we present the case of a patient who arrived in the ED status post suspected alcohol withdrawal seizure with a large, obstructive LH and thrombocytopenia. He required early intubation. The decision to use light sedation allowed him to continue breathing spontaneously with an intact gag reflex while the best airway technique was selected and implemented. Despite reversing the coagulopathy, the LH continued to expand and ultimately resulted in bilateral TMJ dislocation. Until the glossomegaly subsided, the TMJ dislocation was not reducible. The authors suggest this complication might be under-reported, as KN has seen this condition twice, once related to trauma and once due to ACE inhibitor angioedema.

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