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Traumatic lingual hematoma after generalized tonic–clonic seizure in a patient with an acquired coagulopathy

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Abstract:

Oral lacerations are common complications of seizures and account for 92% of all oral injuries. Seizures are relatively commonly associated with chronic alcohol consumption. It is already known that provoked seizures can occur after a sudden cessation of prolonged alcohol intoxication. Meanwhile, chronic alcohol consumption can disrupt the blood coagulation process on several levels. This report aims to present a case of generalized tonic–clonic seizure in a man with chronic alcoholism and acquired coagulopathy who suffered severe tongue injury during a seizure. A 45-year-old man was brought to the emergency department after a first-in-life generalized tonic–clonic seizure. He gave information that he bit his tongue during the seizure. Shortly afterward, the patient had another generalized seizure during which he stopped breathing and was intubated. On admission, the patient was sedated, intubated, and on mechanical ventilation, with no signs of focal neurological deficit. A detailed physical examination revealed massive tongue swelling, which was significantly moved forward. Laboratory tests revealed coagulopathy (INR 2,10) severe thrombocytopenia with a platelet count of $50x10^9$/L. Electrolyte values were in the reference range. According to the maxillofacial surgeon's recommendation, he was treated conservatively, and after 2 weeks, he was clinically stable with a significant reduction of lingual hematoma and without new epileptic events. In our case, decreased platelet count and probable platelet dysfunction associated with chronic alcohol consumption and tongue bite during generalized tonic–clonic seizure played a significant role in developing lingual hematoma. These fast-developing lingual hematomas can lead to possible airway obstruction; therefore, careful observation and timely intubation are mandatory to prevent possible fatal complications.

Keywords:

Coagulopathy, lingual hematoma, provoked seizures

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Introduction

Provoked seizures occur as a consequence of synchronous excessive neural activity due to an underlying cause such as electrolyte disturbance or substance abuse. Some of them are associated with loss of consciousness and falls, which can cause injury in people with epilepsy.[1] Oral lacerations are one of the common complications of seizures and account for 92% of all oral injuries. These are usually minor injuries that require some degree of pain control and are more common in generalized tonic–clonic seizures.[2]

The connection between alcohol abuse and provoked seizures has been already previously known. Alcohol usually acts in the brain as a depressant drug, which in turn increases the seizure threshold. As

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a rebound phenomenon, seizures often occur after a sudden cessation of prolonged intoxication.[3]

Chronic alcohol consumption can disrupt the blood coagulation process on several levels, including thrombocytopenia, thrombocytopathy, or decreased fibrinolysis. These effects can have serious medical consequences, such as increased risk of bleeding.[4]

This report aims to present a case of the first generalized tonic–clonic seizure in a man with chronic alcoholism and acquired coagulopathy who suffered severe tongue injury during a seizure and how this case should be managed in the emergency department.

Case Report

A 45-year-old man was brought to the emergency department after a first generalized tonic–clonic seizure with no known history of previous epilepsy. In the past medical history, the patient had hypertension, chronic alcohol consumption (approximately more than 80 g of alcohol a day), chronic hepatitis B infection, and possible liver cirrhosis (without being fully medically evaluated in the past for the latter two). He denied consuming alcohol in the past 48 h. The patient was aware and oriented, with a normal neurological examination. He also was afebrile and normotensive. He gave information that he bit his tongue during the seizure. The patient denied taking any oral anticoagulants and was not on any antihypertensive drugs (including angiotensin-converting enzyme [ACE] inhibitors). Shortly afterward, the patient had another generalized tonic–clonic seizure during which he stopped breathing and was intubated. He was treated with intravenous (i.v.) benzodiazepines (midazolam 10 mg) and then propofol (100 mg) for the purpose of intubation. The intubation itself was not complicated and was successful on the first attempt. After the seizure was stopped, the patient was admitted to the intensive care unit of the department of emergency neurology. On admission, he was sedated, intubated, and on mechanical ventilation, with no signs of focal neurological deficit. A detailed physical examination after intubation revealed massive swelling of the tongue, which was significantly moved forward [Figure 1] and [Figure 2].

Laboratory tests revealed severe thrombocytopenia with a platelet count of 50 × 10^9/L. The coagulation profile showed elevated prothrombin time (PT) (19.2 s), international normalized ratio (INR) (2.10), and D-dimer (8.16 mg/L). Electrolyte values were in the reference range. Computed tomography of the head and electroencephalogram showed no abnormalities. Conservative management with local cold bandages soaked in adrenaline solution was advised by maxillofacial surgery. i.v. platelet infusion after the peripheral smear analyses was advised by a hematologist for coagulopathy and thrombocytopenia. Further investigations showed no other specific causes of inherited or acquired coagulopathy other than chronic alcohol consumption. For the prevention of new seizures, delirium, and development of Wernicke encephalopathy, diazepam (40 mg) was introduced into the therapeutic regimen alongside thiamin >500 mg i.v. in the first 3 days (followed by a dose of 200 mg/day). The patient was also symptomatically treated for pain with i.v. painkillers (acetaminophen 4 × 1 g/24 h). After 7 days, the patient was extubated, continued spontaneous breathing, and had no new seizures. Repeat laboratory results after i.v. platelet infusion showed recovery of platelet counts to 212 × 10^9/L with similar values of coagulation profile, i.e., PT of 18.4 s and INR of 1.57. Physical examination of the tongue showed a marked reduction of swelling and hematoma. After 2 weeks of hospitalization, the patient was clinically stable with a further reduction of lingual hematoma, and no new epileptic events were observed. Repeat laboratory results before discharge showed platelet levels of 113 × 10^9/L and coagulation profile as PT of 17.9 s and INR of 1.54. The patient was advised to contact infectious diseases specialist because of chronic hepatitis B infection in the history after the end of current hospitalization. Our patient gave informed consent to use his anonymous data in scientific publications.

Figure 1: Massive swelling and hematoma of the tongue caused by bite during a tonic–clonic seizure in a patient with an acquired coagulopathy anterior view

Discussion

Lingual hematoma is a potentially life-threatening phenomenon due to its tendency to cause upper airway obstruction. While tongue bite is a relatively common phenomenon in generalized tonic–clonic seizures, severe tongue swelling and hematoma development are rare complications of seizures. In the previously published work, it is known that bleeding from the

Figure 2: Massive swelling and hematoma of the tongue caused by bite during a tonic–clonic seizure in a patient with an acquired coagulopathy lateral view

lingual artery or its branches can result in very dramatic tongue enlargement. Earlier reports have shown that this enlargement may result in the tongue being displaced in a cephalad and posterior direction endangering the patient's life. Renehan and Morton in their classification of acute enlargement of the tongue have proposed the classification based on the various etiologies encountered.[5]

In our case, the patient had suffered a lingual hematoma secondary to two subclasses of etiology (trauma and coagulopathy), mediated by two generalized tonic–clonic seizures and coagulopathy (that could be caused by alcoholism and possible liver disease with chronic hepatitis B infection). Other possible causes of coagulopathy such as effects of the specific medication (ACE inhibitors and oral anticoagulant therapy) were also considered and as indicated in the case presentation section. Our patient denied taking any specific medications that can have a possible negative effect on coagulation cascade as oral anticoagulants or any antihypertension drugs. We found this as important data in patient's history because Bauriedel *et al*. reported that ACE inhibitors may decrease platelet aggregation by 30% measured by whole-blood aggregometry.[6]

In the cases of acute tongue enlargement, airway management is of prime importance.[6] Previous literature has shown that in some cases, endotracheal intubation is often difficult to perform orally and often must be nasally performed.[7] After the initial airway management, the second critical step in the patient management and further therapy is the identification of the cause of acute tongue enlargement.[7] To make the appropriate medical decision, it is essential to do a careful evaluation of medical history, clinical signs, and laboratory data.[7] Patients who have seizures and are followed up in the postictal period after their seizures are at risk of developing a possibly fatal lingual hematoma if not appropriately monitored. The progressive bleeding on the tongue can cause breathing difficulties due to the trauma during the seizure. These patients are more likely to be unnoticed and may go into respiratory arrest.

Reija *et al*. in their work have stated that blood clotting or coagulation, an important physiological process that ensures the integrity of the vascular system, involves the platelets or thrombocytes as well as several proteins dissolved in the plasma. It is also previously known that thrombocytopenia is a frequent complication of alcoholism, affecting 3%–43% of nonacutely ill, well-nourished alcoholics.[7] It is known that excessive alcohol consumption affects not only platelet production but also platelet function, resulting in a wide spectrum of abnormalities, such as impaired platelet aggregation, decreased secretion or activity of platelet-derived proteins involved in blood clotting, and prolongation of bleeding in the absence of thrombocytopenia.[7]

The management of cases of lingual hematoma greatly varies depending on the cause of the hemorrhage. Previous studies have shown that hematoma evacuation of the tongue is not usually indicated because the bleeding occurs in the intrinsic muscles of the tongue rather than in the potential anatomic fascial spaces; thus, further treatment of lingual hematoma is usually conservative.[7] In our specific case, a maxillofacial surgeon was consulted, and conservative management as already described above in the case presentation was the treatment method of choice.

Conclusion

Lingual hematomas can be a deadly phenomenon requiring rapid identification and management. In our case, decreased platelet count and probable platelet dysfunction associated with chronic alcohol consumption and tongue bite during tonic–clonic seizure played a significant role in the development of lingual hematoma. These fast-developing lingual hematomas can lead to possible airway obstruction; therefore, careful observation of the patient at risk in the postictal period is mandatory to prevent possible fatal complications.

Author contribution statement

BR: Writing – original draft, investigation (equal), visualization. MŽ: Investigation (equal), visualization, writing – review and editing (equal). AR: Supervision (supporting), writing – review and editing (supporting). IB: Conceptualization, supervision (lead), writing – review and editing (equal).

Conflicts of interest

None Declared.

Consent to participate

Our patient gave informed consent to use his anonymous data in scientific publications.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images, and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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