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Cover Page Footnote

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CASE REPORT

Multiple Oral Ulcerations in a Seizure Patient with Undiagnosed Brain Arteriovenous Malformation

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ABSTRACT

Multiple ulcerations are rarely caused by a traumatic event, but seizures can induce repeated injury to the oral mucosa. A patient with a seizure history possesses a challenge in their dental management. **Objective:** In order to describe a case of multiple oral ulceration in a post-seizure patient with undiagnosed arteriovenous malformation (AVM) in the left parietal lobe and provide short guidelines for dentists in managing patient with a seizure history. **Case report:** A 23-years old female had multiple oral ulceration in the right and left buccal mucosa and her tongue. She reported having a seizure attack six days before. The patient never had a history of seizures or recurrent oral ulceration. During the seizure, improper placement of the spoon caused further damage to her oral mucosa. The trigger of her seizure is unknown. Based on the neurological consultation, she had an AVM on the left temporal lobe. In order to relieve pain, mouthwash containing aloe vera, sodium hyaluronate, glycyrrhettinic acid, and polyvinylpyrrolidone (PVP) was prescribed. **Conclusion:** Dentists should be aware that oral mucosal injury can be related to a seizure disorder. A proper history and examination are essential to make an appropriate diagnosis. A safe dental treatment in seizure patients can be delivered accordingly.

Key words: brain arteriovenous malformation, oral ulceration, seizure, traumatic event

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INTRODUCTION

Oral ulceration has many etiological factors, such as infection, immunological disturbance, and traumatic events. Multiple ulceration is commonly related to infection such as intraoral herpes and immunological disturbances, such as recurrent aphthous stomatitis. A traumatic event rarely causes multiple ulcerations, but it is possible as the extent of the lesions depends on the cause of trauma. It is also vital to differentiate multiple ulcers of other etiology.¹⁻³

A seizure is a sign of disease characterized by the discontinuity of normal brain function due to sudden excessive or inadequate electrical discharges. It might cause episodic convulsion or disturbance of movement, disturbance of perception, feeling, or alteration of consciousness.^{4,5} A seizure can occur when there is an imbalance between excitation and inhibition in the brain that resulted from an alteration of brain function, from genes to widespread neuronal circuits.⁶ Globally, about 5% of the population may have a seizure event once in their lives.⁷ Another common term is epilepsy,

which is different from seizure. Epilepsy is a chronic disease that involves seizures and characterizes by changes in perception, behavior, and mental activities.⁴

Patients with seizures have a greater risk of getting an oral mucosal and dental injury.^{8,9} Some patients experience a sudden onset of seizure with unknown provoking factors; thus, they could not prepare safety precautions to prevent injury.⁹ Information regarding seizure disorders and correct recommendations when the seizure happens in the dental office need to be updated. This case report describes multiple oral ulceration in a post-seizure patient with undiagnosed AVM in the left parietal lobe. She had a sudden onset of a seizure, causing inevitable mucosal injury.

CASE REPORT

A 23-years old female, a clinical dental student, had multiple ulcerations in her tongue and cheek, which were present for five days. Upon history taking, she had experienced a seizure attack 6 days before. She bit her tongue during the seizure, and her friend was trying



Figure 1. A traumatic lesion in the right lateral tongue



Figure 2. Multiple traumatic lesions in the left buccal mucosa



Figure 3. Multiple traumatic lesions in the left buccal mucosa

to put a spoon in her mouth, causing more damage to her buccal mucosa. The seizure attack was last for five minutes, followed by a period of unconsciousness approximately for ten minutes. Before the attack, the patient experienced double vision and light dizziness. When the incident happened, the patient was drinking quite a lot of coffee and inhaling a cloud of cigarette smoke from the smoking area of the café. She was not sure whether those were the provoking factors because she used to do the same previously.

The intraoral examination showed multiple ulcerations in the left and right buccal mucosa and the anterior part of the tongue. The ulcerations were shallow, yellowish, irregular, and have a red border (Figures 1–3). It was

painful with VAS 3, and the pain worsened when eating. There was no damage found on teeth or other hard tissues.

The patient never had a seizure attack or major medical condition previously. The seizure was sudden and without any known provoking factors. After regaining consciousness, she was sent to the nearest hospital Emergency Room. MRI examination revealed an arteriovenous malformation at the left parietal lobe. Formerly, she had no related cerebrovascular symptoms.

The oral lesions were diagnosed as traumatic ulcers caused by the seizure attack. Neither the patient nor her family has a history of recurrent oral ulceration. She rarely experienced oral ulceration, and when she has one, it was a single ulcer caused by a known cause such as a toothbrush or removable orthodontic appliance irritation. There was no fever, blister, or other prodromal symptoms before the ulceration. She was not in her menstrual period or under physical or emotional stress when the lesion occurred. No medication was taken for any medical condition.

In order to relieve the pain caused by oral ulcers, the patient was given mouthwash containing aloe vera, sodium hyaluronate, glycyrrhettinic acid, and polyvinylpyrrolidone (PVP) twice a day. The ulcer healed after several days, and the patient never had recurring oral ulcerations afterward. The left parietal lobe arteriovenous malformation was treated via gamma knife surgery by a neurosurgeon. The patient recovered well, and there was no single seizure attack since the surgery. There were no reported cognitive impairments since the surgical treatment. The patient was able to continue her activities as a clinical dental student.

DISCUSSION

A seizure can cause oral soft and hard tissue injury. In this case report, the patient was presented with multiple mucosal injuries associated with seizures with unknown provoking factors. The seizure attack happened suddenly, and no one anticipated the event. Indeed, this history required special attention from dental health professionals as the patient would be susceptible to have another attack. It is a challenge for dental professionals to estimate the risk and readily deliver proper treatment to the patient with a seizure history. This case report will refresh some information about seizure disorder and the dental management of this group of patients.

The dentist's perspective in managing a seizure or epileptic patient may affect their professional interaction with this group of patients.¹⁰ Dentists can administer safe and effective dental treatment to a

patient with a history of seizure. A detailed anamnesis should be obtained before any treatment begins, includes the patient's previous seizure episodes and conditions that provoke epileptic seizures. Dentists should have enough knowledge about the recognition of the early signs of a seizure, a precaution before it occurs, and supportive care if it does occur.⁴

The history of the seizure must be obtained and updated for each visit. For more details, the dentist should ask the patient about the last seizure recurrence time, the medication history (prescribed or self-medicated, taken regularly or not), the consciousness and respiratory state during a seizure, the presence of aura preceding the attack, and the existence of status epilepticus.⁴ By having enough information, dentists can increase their confidence in delivering treatment to this group of patients.

Stress and direct bright light can provoke a seizure. Ensure that treatment is delivered in a short time. It is suggested to make an appointment in the morning. A direct light into the patient's eyes should be avoided.⁴ Elective treatment must be postponed if the patient is known to have more than one seizure per month. In our case, the seizure occurred without warning, catching the patient and surroundings unprepared, which led to her injury. The practice of putting a spoon or any objects in the patient's mouth during a seizure is not recommended as it can result in further injuries.⁸

After consultation with a neurologist, the patient was diagnosed with an arteriovenous malformation in the left parietal lobe. Arteriovenous malformation (AVM) is anomalous communication between arteries and veins resulting from angiogenesis disorders. The pathogenesis is related to abnormal angiogenesis during the embryonic period associated with an action of vascular growth factors.¹¹ Brain AVMs have an estimated incidence of 1:10.000 persons.¹² The most common manifestation is intracranial hemorrhage (ICH) and seizures. AVMs can cause seizures without prior symptomatic ICH. The most common seizure type associated with AVM is generalized seizures.^{13,14} Sometimes, AVM will remain symptomatic and detected by incidental findings in brain imaging.¹⁴

Seizures due to brain AVMs occur in 30–61% of patients; however, the seizure itself is often not the focus of treatment, but seizure control can be attained after complete eradication of the lesion. Based on the meta-analysis cohort by Baranoski et al. (2013), most patients with brain AVM remain seizure-free after proper treatment. The rate of seizure-free outcomes in AVM is superior with microsurgical treatment.¹²

If the seizure happens in the dental office, the most important thing is to prevent injury. Stop all the treatment, and if it is possible to safely do, remove all

intraoral instruments, including dental tampons and removable appliances. Put the patient into a supine position, and there is no need to restrain or move the patient to the floor. It is also not necessary to place any object in the patient's mouth to stop swallowing the tongue. If previously, the patient had an aura, quickly insert a thick gauze tampon to prevent injury to the tooth. Any tight cloth should be loosened.⁴

Make sure the airway is not obstructed.⁴ Turn the patient to the side to prevent any possible aspiration.⁵ After the seizure, some patients will fall into a deep sleep; monitor the patient closely. If the seizure is prolonged even after drug administration, send the patient to the hospital as it can be a sign of status epilepticus, which can be life-threatening.^{4,15} After the patient regain consciousness, make sure the patient is not alone when sent home.⁴

Patients presenting with seizures cannot easily be diagnosed with epilepsy. Several critical steps must be taken before classifying a seizure. The clinician must determine whether the attack was indeed an epileptic seizure or other possibilities of differential diagnoses.¹⁶ Some mimicking diagnoses are syncope, psychogenic seizures, parasomnia, breath-holding spell. It is important to differentiate epileptic from non-epileptic disorder because some conditions may only need reassurance or avoidance of the precipitating factors and do not require antiepileptic medication.⁶

The International League Against Epilepsy (ILAE) stated an operative definition of epilepsy in an official report in 2014. Epilepsy is considered when someone has any of the following conditions: at least two or more seizures occurring more than 24 hours apart, one unprovoked (or reflex) seizure, and a probability of further seizures similar to the general recurrence risk (at least 60%) after two unprovoked seizures, occurring over the next 10 years, and have a diagnosis of an epilepsy syndrome.^{17,18}

As a dentist, it is important to diagnosed multiple oral lesions correctly as it has many differential diagnoses, and each has a different treatment plan. In this case, the patient was present with shallow, irregular, yellowish multiple ulcerations, but there was no history of recurrent oral ulceration. No family member has recurrent oral ulceration, and the patient only experiences oral ulcers with known causes. The patient also did not experience fever or blister preceding the ulcer, as seen in the prodromal stage of recurrent intraoral herpes. Furthermore, the lesions are located at nonkeratinized mucosa; meanwhile, herpes simplex infection tends to occur in keratinized mucosa.² The differential diagnosis of recurrent aphthous stomatitis and recurrent intraoral herpes can be excluded. Traumatic ulcers rarely have multiple presentations, but the cause of the ulcerations was clearly due to soft tissue injuries caused by surrounding teeth during the seizure

attack. The patient reported no recurring ulceration after the seizure attack. Patients with traumatic ulceration eventually will heal within several days, but it is important to relieve the pain and boost healing.¹ Pain in the orofacial area, which makes the patient uncomfortable, might provoke seizures. It is necessary to eliminate the pain before complications arise.⁴

In this case, we use a mouthwash that containing aloe vera, sodium hyaluronate, glycyrrhettinic acid dan polyvinylpyrrolidone (PVP). Aloe vera is capable of relieving pain and erythema, reducing lesion size, and promote the healing of oral ulcers in some studies.¹⁹ Hyaluronic acid (as sodium hyaluronate) acts as a coating agent in oral mucosa, enhances tissue hydration, and accelerates healing.²⁰ Many studies have shown that hyaluronic acid application can improve ulcer healing in subjective parameters (e.g., by reducing pain and shorten the period of healing) and objective parameters (ulcer size, ulcer numbers, and inflammatory signs).^{21,22} Polyvinylpyrrolidone has muco-adherent and film-forming properties that can enhance tissue hydration. Glycyrrhettinic acid has anti-inflammatory properties that help in ulcer healing.²⁰

It is important to know the oral healthcare need of people with seizures or epilepsy. A study from Karolhazy shows that patients with epilepsy have poor oral hygiene practices, injuries to the oral cavity, and increased force on the teeth due to the seizure. Furthermore, dental treatments were not adequately delivered to such patients. Insufficient knowledge about this disorder and the fear of a seizure attack during dental treatment might be the root of this attitude.²³

Oral lesions and seizures are rarely described in the literature; most injuries described are related to dental or jaw injury. Patients with a seizure disorder are prone to get a crown fracture in the upper central incisor, as shown in a study by Gerreth (2014).⁸ Minor oral cavity injuries are often caused by generalized tonic-clonic seizures as the patient bites the tongue or other areas of the oral mucosa. Frequently it also leads to dental injuries because of the fall or the forceful contraction of the masticatory muscles during both the tonic and clonic phases.²³

Another soft tissue disturbance related to seizure is gingival overgrowth related to the antiepileptic drug, especially phenytoin. About half of the patients taking phenytoin will develop gingival hypertrophy within 1–2 years after the initiation of treatment, but it will recede within months after discontinuation.^{23,24} Good oral hygiene practices can prevent or improve the condition.²⁴

If the cause of oral soft tissue injury is identified correctly, the right treatment can be delivered. The lesion severity should be evaluated, and the next possible recurrence should be prevented. The management of oral lesions in a patient with a history

of seizures is thoroughly challenging since the seizures are unpredictable. The dental team must be ready for any possible occurrence at the dental office. Dentists must be knowledgeable in order to deliver safe dental treatment. Thus, the dentist's ability to identify the risk and severity of a neuromuscular disorder by getting thorough medical history is vital.

CONCLUSION

Dentists should be aware of any oral injury related to seizure attacks and enhance their knowledge to improve oral health for this group of patients. A profound anamnesis related to any signs and symptoms, which lead to seizure disorder is important to help identify and estimate the severity of the disease. The management of patients with a serious seizure disorder should be done in collaboration with a neurologist or neurosurgeon. With the proper understanding of the disease, oral healthcare professionals can safely deliver comprehensive oral health care for patients with a seizure history.

CONFLICT OF INTEREST

The authors declare there is no conflict of interest.

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