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Consider a new diagnosis of epilepsy as a cause of tongue ulceration

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Abstract

Background: Tongue ulceration is a common presentation to dental practitioners. The most likely causes are: aphthous stomatitis, geographic tongue, trauma, or autoimmune conditions such as lichen planus (Byahatti and Ingafou, 2010). We report a patient who presented with recurrent tongue ulceration in whom epilepsy was not considered as the cause until a seizure occurred while awake, months later.

Conclusion: Epilepsy should be considered as a differential diagnosis of unexplained traumatic lateral tongue ulceration.

Case History

A 61-year-old female (SB) was referred to a dental specialist clinic for assessment of sudden onset lesions of the tongue, emerging exclusively at night. The patient thought there was a temporal relationship with eating fish for dinner. There were no obvious mucosal lesions at her first visit. No treatment was prescribed, and she was asked to return with any recurrence.

At the next visit the patient provided photographs of her tongue showing possible bilateral purpura or haemorrhagic bullae of the tongue. On examination of her tongue, there was again no sign of ulceration.

SB had normal oral health prior to this, with no history of oral ulceration, easy bruising or bleeding. Her medical history included osteoarthritis and unconfirmed cutaneous lichen planus. The ulcers were sore when present but there was no other significant pain history. Peripheral examination revealed no active erythema or oedema of the hand joints and no nail dyskeratosis. The lower lip had a morsicatio pattern internally and she had prominent bilateral linea alba. There was a few reticular striations of the left posterior buccal mucosa. Focal fibrous hyperplasia was noted on her lateral tongue bilaterally. General haematological investigations were also undertaken and found to be within normal limits. There was a slight rise in antinuclear antibody level.

The differential diagnoses included trauma from a parafunctional tongue biting habit, a food related mucosal allergy, a leukocytoclastic vasculitis or a vesiculobullous disorder. It was considered unlikely that oral lichen planus was a contributing factor. The patient was provided with firm maxillary acrylic bite splint (Michigan type) to wear at night, and to return for biopsies should ulceration recur.

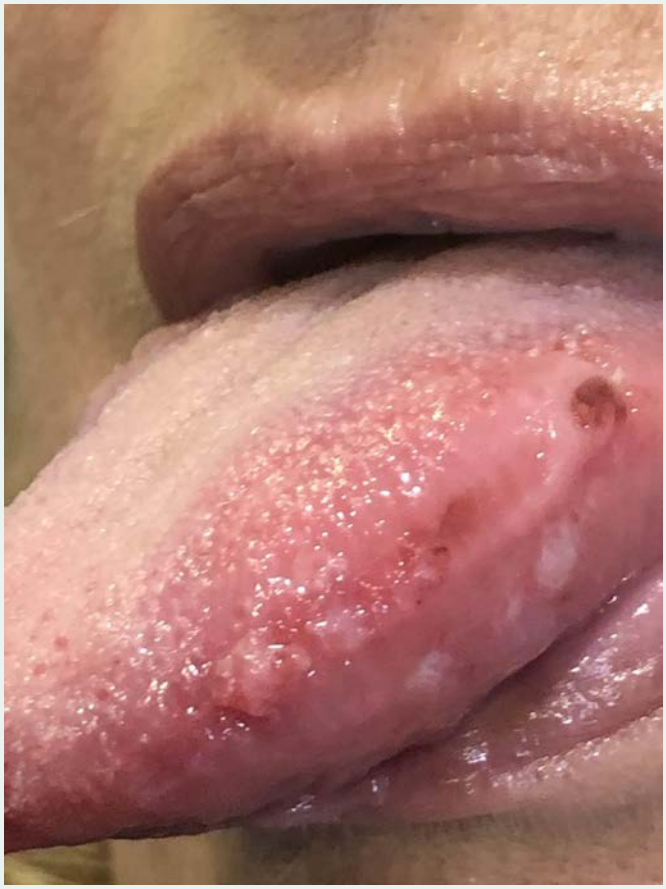

Figure 1. Extra-oral photo of the left side of the tongue following an event. Areas of ulceration and hyperplasia are visible along the lateral border, within a large, erythematous region.

Later, as directed, she presented acutely with the sudden onset of a new tongue ulceration (Figure 1). Her original dental specialist was not in the clinic that day. An alternative dental specialist thought the lesions looked traumatic in origin and as requested took biopsies to exclude leukocytoclastic or vesiculobullous disorders. The biopsy results were consistent with trauma. The patient was then referred onto a third dental specialist to consider making a bimaxillary device like an oral snoring appliance to make it impossible for her to bite the lateral surfaces of her tongue.

Three months following her final consultation, SB woke up on her kitchen floor at 4.15 pm. Her last memory was of cleaning the oven at 3.15 pm, without any warning to a loss of consciousness. Subsequent neurological consultation did not identify additional symptoms to indicate mesial temporal epilepsy, such as déjà vu sensations or other seizure semiology. There had been

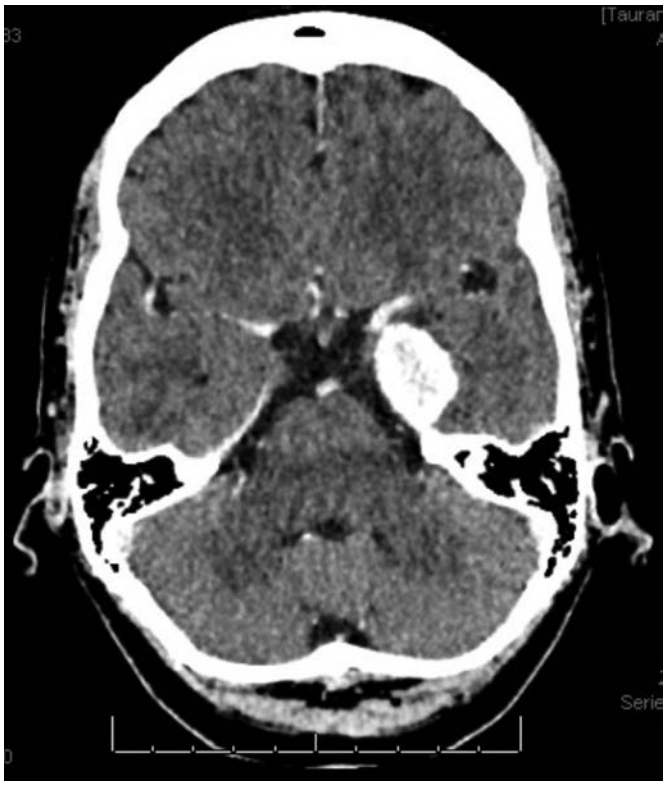

Figure 2. CT brain post contrast with 4 cm partially calcified enhancing mass typical of a meningioma, arising from the left cavernous sinus region and tentorium cerebelli with mass effect on the adjacent left temporal lobe.

no complications of tonic clonic seizures, such as widespread myalgia, other than tongue trauma with the nocturnal events.

Further investigations revealed a meningioma (Figure 2); SB was commenced on the anti-epileptic drug (AED) Lamotrigine and referred for neurosurgery. A left pterional craniotomy with total excision of the tumour was complicated by a few months of diplopia, consistent with a fourth cranial nerve palsy, for which she wore a prism in her spectacles. Tumour histology was consistent with a WHO Grade I meningioma, with some foci of necrosis. A post-operative MRI showed total removal of the tumour.

Since treatment commenced, SB has no tongue ulceration or any other evidence of a seizure. The patient remains on AED treatment and is ineligible to drive until a year has elapsed from the last seizure.

Discussion

Seizures and epilepsy are paroxysmal disorders of the brain with many causes and variable semiology characterized by either focal, or generalised discharges of dysfunctional neurons which may be recorded with electroencephalography (EEG). Orofacial trauma is a common complication, with injury to the lips, tongue, teeth or face (Falci et al., 2019).

A small minority of all epilepsies are due to brain tumours. Epileptogenesis is influenced by both tumour pathology and anatomical location. For example, low grade astrocytomas and metastatic disease of the brain are more likely to cause epilepsy than meningiomas; while temporal lobe pathologies, intrinsic, or extrinsic (as in our case), are more epileptogenic than other cortical areas (van Breemen et al., 2007).

The sleep-wake cycle has a strong influence on seizure occurrence in many epilepsy syndromes. Indeed, in some patients, seizures may be exclusively nocturnal (Husain and Sinha, 2011). In a patient who has no warning (aura) with amnesia, it is the family or bed-partner that alert medical practitioners from such clues as noisy obstructed breathing, or by direct observation of a convulsion. Some patients will experience symptoms in the pre-ictal or post-ictal phase, but fail to appreciate their significance until after the diagnosis is made.

Diverse injuries are more likely with major (tonic-clonic) seizures than focal onset seizures and include: consequences of motor vehicle accidents, head and facial injuries, cervical fractures, shoulder dislocation, burns, loss of teeth, and commonly, a painful tongue injury due to the powerful contraction of the mastication muscles in the unconscious state. Seizures may be fatal due to injury or drowning, as well as from status epilepticus or sudden unexpected death, usually during sleep (Brennan et al., 2020).

Tongue trauma, of the lateral borders of the tongue, is a specific sign of a tonic or tonic-clonic (convulsive) seizure (Benbadis et al., 1995). Anterior, tongue-tip trauma is less specific and can be due to non-epilepsy related loss of consciousness such as syncope of various causes, or psychogenic (dissociative) non-epileptic seizures (Smith, 2001).

Incredibly, as described to her neurologist after the diagnosis became apparent, SB had experienced more than 30 episodes over several months, of waking before 2.30 am to find blood over her pillow, hair, mouth, neck; even splashed onto the headboard. This was accompanied by a painful tongue but no other features of a tonic-clonic convulsion, such as widespread myalgia, confusion or incontinence.

In our case, the ulceration was established as traumatic but was still attributed to a tongue biting habit. While parafunctional biting habits are common on the cheeks and to a lesser extent the tongue, they are generally associated with a less severe presentation.

Nocturnal bruxism with tongue thrusting is more likely to present as hyperkeratosis or indentations of the tongue as opposed to ulceration or laceration (Murali et al., 2015).

Once the patient was recognised to have epilepsy the cause of the nocturnal tongue ulceration was realised. Many factors conspired to prevent an earlier diagnosis. Tongue ulceration due to epilepsy is uncommon as the only clinical feature. The patient suggested an unlikely explanation of a food allergy. The clinician who finally confirmed the diagnosis of trauma was not the lead clinician who had taken the initial history but happened to be working on the day she presented with the tongue ulceration for biopsy. These factors contributed to cognitive biases.

Conclusion

This case serves as a salutary reminder to dental practitioners to consider a seizure as one cause of tongue ulceration, with the opportunity to contribute to a novel diagnosis of epilepsy.

While nocturnal tongue biting and thrusting habits may result in lesions to the tongue, these mechanisms tend to present in a manner suggestive of chronic irritation. Traumatic ulceration of the tongue heals within days, whereas ulceration from inflammatory mucosal conditions such as lichen planus or vesiculobullous disorders leave lasting effects in the mouth.

Trauma to the lateral tongue is a dependable sign of a convulsive seizure and should prompt a neurology opinion. This said, even in expert hands, the diagnosis of epilepsy is not straightforward and relies heavily on the history and observations from a witness when the patient cannot describe some, or all of the attack.

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References

Benbadis SR, Wolgamuth BR, Goren H (1995). Value of Tongue Biting in the Diagnosis of Seizures. *Arch Intern Med* 155: 2346-2349.

Brennan M, Scott S, Bergin P (2020). Sudden unexpected death in epilepsy (SUDEP) in New Zealand; a retrospective review. *NZ Med J* 133: 65-71.

Byahatti SM, Ingafou MSH (2010). The Prevalence of Tongue Lesions in Libyan Adult Patients. *J Clin Exp Dent* 2: 163-168.

Falci SGM, Duarte-Rodrigues L, Primo-Miranda EF, Gonçalves PF, Galvão EL (2019). Association between epilepsy and oral maxillofacial trauma: A systematic review and meta-analysis. *Spec Care Dentist* 39: 362-374.

Husain AM, Sinha SR (2011). Nocturnal Epilepsy in Adults. *J Clin Neurophysiol* 28: 141-145.

Murali RV, Rangarajan P, Mounissamy A (2015). Bruxism: Conceptual discussion and review. *J Pharm Bioallied Sci* 7: S265-S270.

Smith PEM (2001). If it's not epilepsy... *J of Neurol Neurosurg and Psychiatry* 70: ii9-ii14.

van Breemen MSM, Wilms EB, Vecht CJ (2007). Epilepsy in patients with brain tumours: epidemiology, mechanisms, and management. *Lancet Neurol* 6: 421-430.