Stomatologija. Baltic Dental and Maxillofacial Journal, 27: 20-4, 2025

Post-traumatic oral necrosis in a patient with refractory epilepsy: A case report

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SUMMARY

Epilepsy patients can show a wide range of oral and maxillofacial injuries after seizures. However, the occurrence of oral soft tissue necrosis (OSTN) has been rarely reported. In the current report, a 55-year-old man with refractory epilepsy was referred for evaluation of an extensive injury in the oral mucosa after a severe seizure. Intraoral examination revealed a painless grayish-white necrotic area in the posterior region of the buccal mucosa on the left side measuring about 2×3 cm. Clinical findings led to the diagnosis of post-traumatic OSTN and the patient underwent surgical debridement on an outpatient basis. Two-month follow-up showed complete healing associated with the formation of fibrous tissue. A review of the literature demonstrated that post-traumatic OSTN was not reported among oral and/or maxillofacial injuries found in epilepsy patients. However, the current report reinforces that soft tissue necrosis may occur in the oral cavity of these patients after seizures. Post-traumatic OSTN should be considered an oral injury found in patients with refractory epilepsy.

Keywords: epilepsy, maxillofacial injuries, oral injuries, necrosis, oral soft tissue necrosis.

INTRODUCTION

Epilepsy is a progressive, chronic neurological disease characterized by recurrent seizures resulting from excessive and/or synchronous neuronal electrical discharges (1, 2). The epileptic seizures can manifest as an absence of awareness, sensory changes and/or uncoordinated movements (1, 2). Electrical discharges in brain circuits may have focal-onset and spread throughout the central nervous system leading to severe disturbances of consciousness (3).

Epilepsy patients can be treated with anti-epileptic drugs (AEDs), diet ketogenic or neurosurgery depending on the type of seizure (4, 5). However, about 30% of epilepsy patients do not obtain adequate seizure control presenting refractory epilepsy (4, 5). The lack of an effective control over seizures can lead to the development of cognitive, motor, psychological and/or social disorders (3, 6). Furthermore, these patients are exposed to an increased risk for accidental injuries compared to non-epileptic patients (3, 6).

Due to recurrent seizures, patients with refractory epilepsy often present oral and maxillofacial injuries, such as dental trauma and soft tissue injuries (7-21). However, the occurrence of extensive tissue necrosis in the oral cavity of these patients, associated with a trauma derived from an epileptic seizure, has not yet been reported. In the current report, we describe an unusual case of post-traumatic oral soft tissue necrosis (OSTN) resulting from an epileptic seizure. A review of the literature was performed to identify oral and/or maxillofacial injuries found in epilepsy patients.

CASE REPORT

A 55-year-old white man was referred to our service for evaluation of an extensive lesion in the oral mucosa. According to the patient, it appeared after a severe epileptic seizure and had two weeks of evolution. The patient had a medical history of hypertension and refractory epilepsy and was taken

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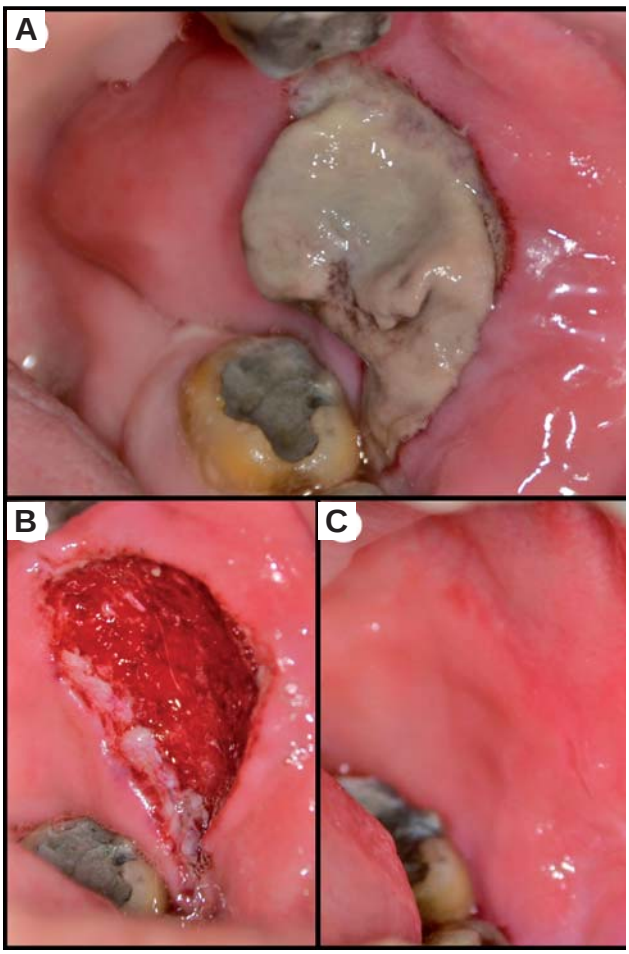

Fig. A – extensive post-traumatic necrosis in the left buccal mucosa of a 55-year-old male patient with refractory epilepsy. B – surgical wound after debridement of the necrotic tissue. C – complete healing associated with the formation of fibrous tissue after two months of follow-up.

hydrochlorothiazide (25 mg), losartan potassium (50 mg), phenytoin (400 mg) and phenobarbital (300 mg) daily. Complementary exams did not reveal changes in blood cell count and glucose levels.

Extraoral examination showed no abnormalities. Intraoral examination revealed a painless grayish-white necrotic area in the posterior region of the buccal mucosa on the left side measuring about 2 x 3 cm (Figure, A). Second molars were occluding close to the lesion suggesting their involvement with the tissue damage. Clinical findings led to the diagnosis of post-traumatic OSTN. Surgical debridement was performed on an outpatient basis (Figure, B) and the histopathological analysis revealed a necrosis. The patient was followed-up weekly for postoperative control. A two-month follow-up showed complete mucosal healing associated with the formation of fibrous tissue (Figure, C). The patient is being monitored in our service and did not present other oral injuries related to epileptic seizures after two years of follow-up.

A review of the literature was performed to verify the occurrence and types of oral and/or maxillofacial injuries found in epilepsy patients. The current review included observational studies, published in English between 2000 and 2021 and was conducted in PubMed/Medline using a combination of the following terms: “Maxillofacial trauma” or “Maxillofacial injury” or “Orofacial trauma” or “Orofacial injury” or “Oral cavity trauma” or “Oral cavity injury” or “Oral trauma” or “Oral injury” and “Epilepsy” or “Seizures”. Research articles published in other languages, literature reviews and case reports were excluded from this review. The results of the current literature review are shown in Table.

DISCUSSION

Studies have shown that patients with refractory epilepsy may exhibit an increased number of oral and maxillofacial injuries than patients with controlled epileptic seizures (13, 17, 19, 21). Most of these injuries occur during daily activities at home or work and are usually associated with biting or forced insertion of objects into the mouth during crises (7, 13-15, 19). In the current clinical report, the patient showed a history of refractory epilepsy and would have bitten the buccal mucosa during the last seizure. This accidental trauma caused the rupture of blood vessels and tissue devascularization. Both events led to the formation of an unusual extensive area of soft tissue necrosis in the oral mucosa, not previously reported in epilepsy patients.

According to the literature review, a total of 3.289 patients displayed epilepsy. Of these, 801 (24.35%) had at least one oral or maxillofacial injury (7-21). Dental trauma was reported in all studies included in the current review and comprised fractures (7, 14-16, 18, 20), avulsion (8, 11, 12, 16) and luxation (11, 12). In most cases, the incisors were affected during the seizures (8-10, 15). Lacerations (7, 11, 14, 16-18, 20) and bruises (7, 18) were the most common soft tissue injuries found in these patients. Both occurred more often in lips (7, 14, 16, 18, 20) and tongue (7, 13, 14, 16, 18). No studies included in the review reported the occurrence of post-traumatic OSTNs related to epileptic seizures. The occurrence of mandibular fractures (11, 12) and temporomandibular joint luxation (11) was also reported in epilepsy patients.

In the current report, we showed the occurrence of a post-traumatic OSTN in a patient with refractory epilepsy. Herein, the injury was caused by a mechanical trauma that occurred during the epileptic seizure and the diagnosis was established according to the clinical findings. Clinical manifestations of the post-traumatic OSTN usually include yellowish-white tis-

sue, which can be associated with edema, erythema, ulcer and/or pain (22). In addition to mechanical trauma, chemical substances and radiotherapy may also promote tissue necrosis in the oral cavity (24-28). However, these cases often do not show extensive areas of necrotic tissue, which are found in cases of post-traumatic origin (22-28). A high-intensity trauma may lead to the rupture of large blood vessels that nourish the oral mucosa resulting in an OSTN (29).

Most cases of OSTN are managed conservatively (23-28). The areas of superficial necrosis usually do not need specific treatment, resolving completely within two weeks (23-28). However, surgical debridement is required for the treatment of extensive areas of necrotic tissue (22, 30). In the current clinical report, all necrotic tissue was removed to improve microcirculation as well as stimulate the healing of the buccal mucosa. Surgical debridement also avoids

Table. The focus question development according to the PICOS study design

Author, year	Study design	Epilepsy patients	Non-epileptic	Population	Number of oral and maxillofacial injuries	Type of injury
Appleton *et al.*, 2002	Cross-sectional	198	0	Children with newly diagnosed and untreated epilepsy	25	Dental trauma and oral soft tissue injuries
Percival *et al.*, 2009	Case-control	39	39	EP: children and adolescents with intractable epilepsy NE: healthy children	EP:40 NE: 4	Dental trauma
Thomas *et al.*, 2009	Cross-sectional	1673	0	NS	14	Dental trauma
Gurbuz *et al.*, 2010	Case-control	211	211	EP: NS NE: healthy individuals	EP: 58 NE: NS	Dental trauma and facial trauma
Nonato *et al.*, 2011	Case-control	159	68	EP: NS NE: healthy individuals	EP: 79 NE:3	Dental trauma, oral soft tissue injuries and temporomandibular disorders
Costa *et al.* 2011	Cross-sectional	109	0	Patients treated for refractory epilepsy	89	Dental trauma and facial trauma
Adewole *et al.* 2011	Cross-sectional	138	0	NS	87	Dental trauma and oral soft tissue injuries
Mollaoğlu *et al.*, 2013	Cross-sectional	126	0	NS	61	Dental trauma and oral soft tissue injuries
Gerreth *et al.*, 2014	Case-control	63	63	EP: epilepsy students NE: healthy students	EP: 10 NE: 4	Dental trauma
Ghafoor *et al.*, 2014	Cross-sectional	150	0	Epilepsy children	222	Dental trauma and oral soft tissue injuries
Lagunju *et al.*, 2015	Case-control	125	125	EP: epilepsy children NE: healthy children	EP:57 NE:26	Dental trauma and oral soft tissue injuries
Gawlak *et al.*, 2017	Cross-sectional	106	0	NS	55	Dental trauma and oral soft tissue injuries
Cengiz *et al.*, 2019	Cross-sectional	57	0	NS	13	Dental trauma
Morgan *et al.*, 2019	Case-control	100	80	EP: epilepsy children NE: healthy children	EP: 7 NE: 0	Dental trauma and oral soft tissue injuries
Yeung *et al.*, 2019	Case-control	35	35	EP: epilepsy children NE: healthy children	EP: 8 NE: 8	Dental trauma and oral soft tissue injuries

EP – epilepsy patients; NE – non-epileptic; NS – not specified.

the increase of the tissue necrosis (22, 30).

CONCLUSION

The current report reinforces that soft tissue necrosis may occur in the oral cavity of patients with refractory epilepsy after mechanical traumas derived from seizures. Post-traumatic OSTN should be included in the wide range of oral and/or maxillofacial injuries related to epileptic seizures.

ETHICAL APPROVAL

This article complies with the ethical standards as outlined by the publisher.

STATEMENT OF CONFLICT OF INTEREST

The authors state no conflict of interest.

FUNDING

The current report did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

ACKNOWLEDGMENTS

LMA and TFC are supported by the Coordenação de Aperfeiçoamento de Pessoal de Nível Superior - Brasil (CAPES) – Financial Code 001.

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Received: 08 09 2022
Accepted for publishing: 24 03 2025