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Tooth-brushing epilepsy: an SEEG study and surgical treatment

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ABSTRACT – We report a patient with reflex tooth-brushing-triggered epilepsy, associated with a post-central lesion within the right somatosensory face area. Contralateral facial sensory and motor phenomena, associated with contralateral upper limb extension, were present at seizure onset after gingival stimulation, but seizures could also be induced by contact with solid food or liquids. Spontaneous seizures also were recorded. Secondary generalization was infrequent. Stereoelectroencephalography implantation was performed, with seizure recording and cortical/subcortical stimulation for mapping, to identify the precise extent of surgical resection. Complete postoperative control of epilepsy was achieved, accompanied by a mild and transient neurological deficit. [*Published with video sequence*].

Key words: tooth-brushing induced seizure, reflex seizures, facial somatosensory and motor areas

VIDEO ONLINE

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Reflex epilepsy is a very rare condition in which seizures are triggered by an external stimulus or internal mental process (Xue and Ritaccio, 2006; Wolf, 2017; Okudan and Ozkara, 2018) Tooth-brushing epilepsy is an unusual form of reflex epilepsy provoked by the act of tooth-brushing, or sometimes by stimulation of the gingival region with liquids or solids. We identified reports from as early as 1891, describing the relationship between caries and epilepsy, indicating that an inflammatory stimulus may affect the lower trigeminal branches and thus provoke seizures (Badowski, 1891). This epileptic phenomenon could originate from lesions located mostly in the central area, as reported elsewhere. (Chan *et al*., 2016).

Case study

We present a 27-year-old, right-handed woman who developed epilepsy at four years of age, with seizures initially occurring during

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sleep. Shortly afterwards, seizures began to be triggered predominantly by tooth-brushing, although spontaneous seizures continued to occur. The presence of food and/or liquid in contact with the oral mucosa or gingival region could also induce seizures. There was no focal neurological deficit.

At the time of the first consultation with our team, the patient was 27 years old, and she identified that more than 80% of her seizures occurred when she brushed her left upper gums.

She had previously received treatment with multiple antiepileptic drugs, including carbamazepine, phenytoin, vigabatrin, lamotrigine, valproic acid, levetiracetam, clobazam, and cannabidiol, as monotherapy or in different combinations, without achieving seizure control. The maximum period of seizure freedom was one week, and she had an average of four to five seizures per day, despite medication.

Her condition had previously been considered psychogenic by others and several brain magnetic resonance imaging (MRI) studies had been interpreted as normal. The patient had a poor quality of life, considering herself severely disabled. A thorough evaluation of a new brain MRI by our neuroradiologist demonstrated a cortico-subcortical lesion in the lower third of the postcentral gyrus (*figures 1, 2*), with blurring of the grey-white matter junction, which could be interpreted as concordant with seizure triggering and semiology.

During preoperative video-telemetry monitoring, right frontal interictal epileptiform activity was present. Two typical tooth-brushing seizures were recorded *(see video sequence)*, in which the patient, after beginning the action of brushing the left upper and lower gums, manifested a left hemifacial tingling sensation located around the lips. Immediately following this sensation, she exhibited left hemifacial tonic contraction, with flexion and then tonic extension of the left arm, followed by clonic contraction of the neck muscles. There was no alteration of consciousness. Ictal EEG demonstrated epileptiform discharges located in the right frontal region, but interpretation was difficult because of the amount of muscular artefact.

Based on seizure semiology, comprised of sensory manifestation on the left face almost synchronically with motor manifestations affecting the left face and upper limb, stereoelectroencephalography (SEEG) implantation was proposed to delineate the extent of the primary epileptogenic area and probable cortical resection. Six depth electrodes were implanted (Depthalon, PMT Corporation, Chanhasset, Minnesota, USA) using the CRW stereotactic frame (Integra, Plainsboro, NJ, USA) *(see video sequence)*.

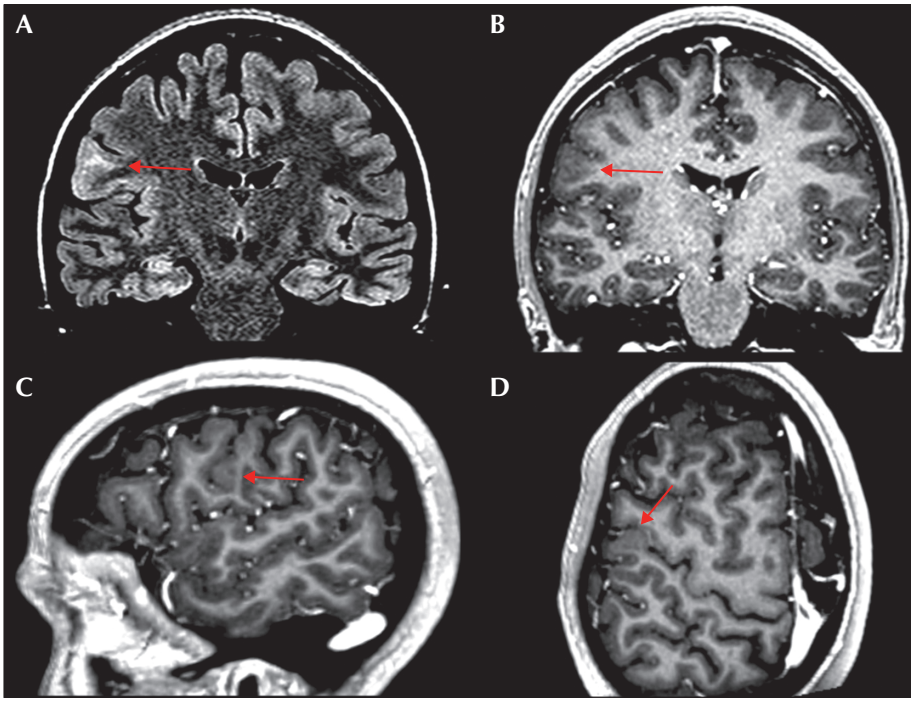

Figure 1. MRI showing a cortico-subcortical lesion in the lower third of the postcentral gyrus (A-D), with blurring of the grey-white matter junction (coronal FLAIR sequence [A]; post-gadolinium coronal T1WI [B]) and subcortical hyperintensity (A). Sagittal (C) and oblique axial (D) post-gadolinium T1WI reconstructions also exhibit the lesion.

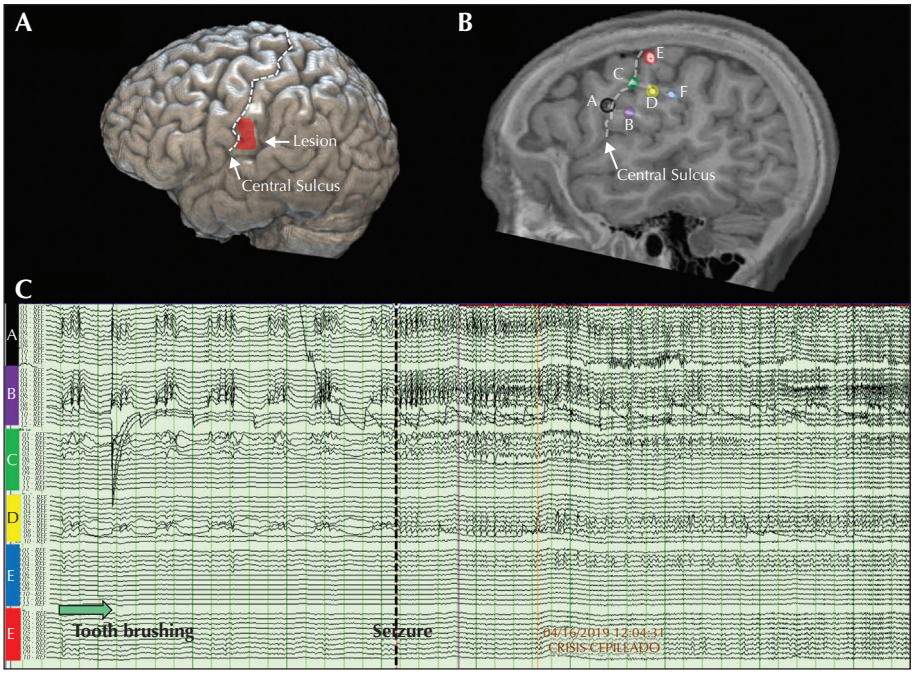

Figure 2. Anatomical relationship between a 3D reconstruction of the patient's brain (A), the location of the electrodes in the sagittal plane in relation to the central sulcus (B), and the ictal trace of the SEEG (C).

These included one in the lower third of the precentral region and frontal opercula (electrode A); three in the lower half of the postcentral gyrus, including the parietal opercula (from inferior to superior: electrodes B, C and D); one in the superior part of the postcentral gyrus at the region of the hand somatosensory area (electrode E); and one in the anterior aspect of the supramarginal gyrus (electrode F) (*figure 2*). Interictal recording demonstrated epileptic activity predominantly in the postcentral and parietal operculum electrodes (B and C), while the precentral electrode (A) also exhibited abnormal activity at the most superficial contacts. During SEEG, four seizures were induced when the patient brushed her teeth, predominantly as she brushed the upper left gingival region. The ictal phenomena exhibited the same sequence as described during preoperative video-telemetry. Seizures lasted approximately 13 seconds. The electrodes involved at the beginning of the seizures were A, B and C (*figure 2*). The patient had no loss of consciousness or post-ictal status and could respond to simple commands immediately during the post-ictal phase. Electrical stimulation was performed, allowing us to map the face sensory (electrode D) and motor area (electrode A) (*see video sequence*). No clinical response was elicited when stimulating the tissue around electrode B, which we interpreted as coincident with the location of the dysplastic lesion on MRI. These findings are consistent with those from Marusic *et al.* (2002), in which no motor functions were found in perirolandic areas with FLAIR MRI signal increase. The descending fibres of the left upper limb within the corticospinal tract were also mapped during stimulation, to define the depth of resection. The anatomical location of the contacts was used for neuronavigational surgical planning.

Resection of the lower third of the postcentral gyrus, the anterior and external part of the parietal operculum, and a portion of the precentral gyrus was performed (*figure 3*). Postoperatively, the patient presented with mild weakness of the lower left side of the face and mild deviation of the tongue towards the left (*see video sequence*). These symptoms were transient and reversed completely within three weeks, as expected from previous experience (Lehman *et al.*, 1994). Histology revealed cortical dysplasia type IIB in the post-rolandic tissue samples, but no histological abnormalities were observed in the pre-rolandic (motor area) samples. Postoperative MRI revealed the performed resection with preservation of the underlying white matter tracts (*figure 4*). Seizure freedom was achieved, with progressive tapering of the patient's medication still on-going.

Discussion

Epileptic seizures are generally unpredictable and arise spontaneously. Patients often report non-specific triggers, such as stress or sleep deprivation; but only rarely do seizures occur as a reflex event, in which they are objectively and consistently precipitated, or

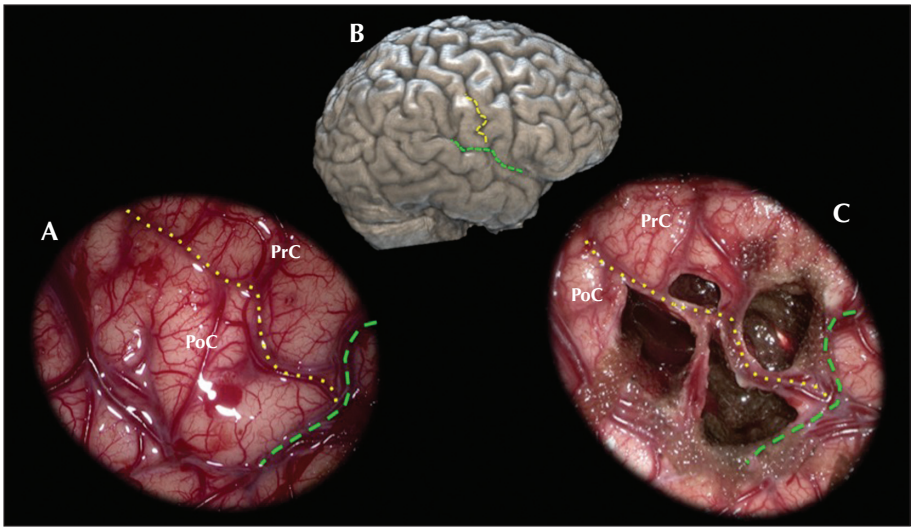

Figure 3. (A) Pre-resection image of the brain showing the postcentral gyrus, precentral gyrus, central sulcus and sylvian fissure. (B) 3D reconstruction of the patient's brain. (C) Post resection image of the brain. PoC: postcentral gyrus; PrC: precentral gyrus; yellow dotted line: central sulcus; green dotted line: Sylvian fissure.

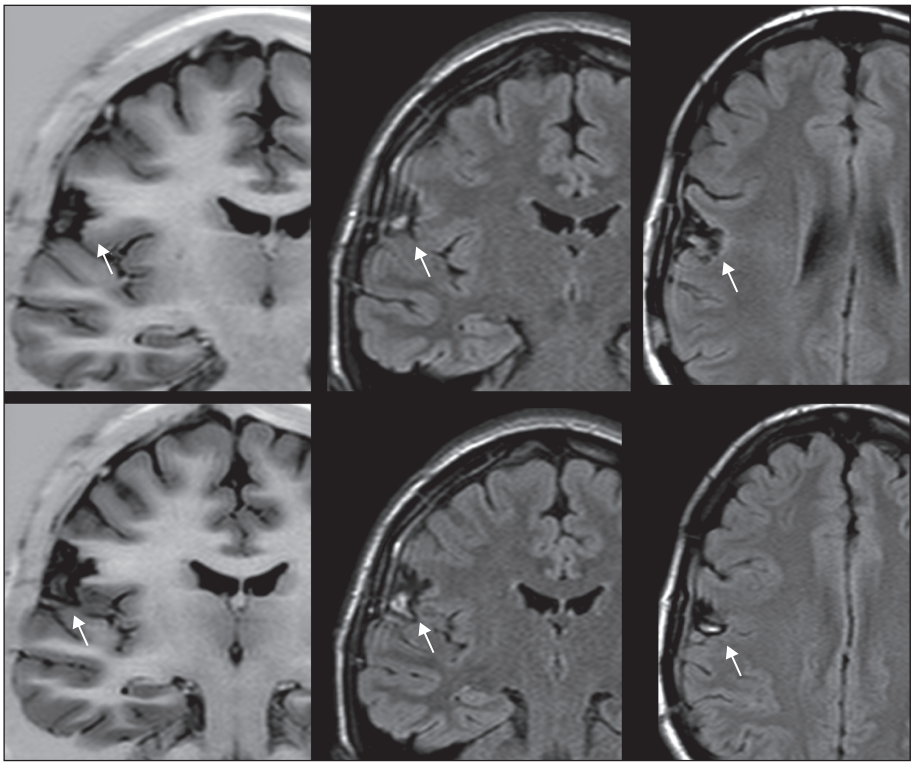

Figure 4. Postoperative MRI showing the cortical resection (white arrow) on coronal plane (inversion recovery and FLAIR sequences) as well as on axial plane (FLAIR sequence).

inhibited by internal or external stimuli (Okudan and Ozkara, 2018; Randhawa *et al*., 2019).

The 1989 International League Against Epilepsy (ILAE) classification established the definition of epilepsy with specific stimuli (Epilepsy, 1989). In 2001, an ILAE task force constructed a list of accepted precipitating factors for reflex seizures (Engel, 2001).

Reflex seizures are an ideal model to study the dynamics of seizure initiation and termination. Tooth-brushing epilepsy is a rare type of reflex epilepsy, and its aetiology is not well established. The periodontal ligament and gingiva represent a highly-innervated zone, although this may require excitation from a critical mass of a previously hyper-excitable

region within the somatosensory cortex to generate a seizure.

The first case of tooth-brushing-induced epilepsy described in the literature was published in 1982, in a 12-year-old boy. Seizures began after placement of a palatal expansion appliance, with right-side Jacksonian seizures evoked by tooth-brushing. The patient's neuroimaging was normal, but electroencephalography detected left frontocentral spikes and the patient failed to respond to antiepileptic drugs (Holmes *et al*., 1982). We have located 17 cases of tooth-brushing-induced seizures described in the literature, but none of them were studied with SEEG recording and/or stimulation for brain mapping (Holmes *et al*., 1982; O'Brien *et al*., 1996; Koutroumanidis *et al*., 2001; Chuang *et al*., 2004; Navarro *et al*., 2006; D'Souza *et al*., 2007; Haytac *et al*., 2008; Kumada *et al*., 2013; Ott *et al*., 2014; Chan *et al*., 2016).

Tooth-brushing seizures are caused by sensitive stimuli in the teeth and gums during brushing. However, the trigger does not appear to be merely a "simple" stimulus. Repetition of the stimulus, and frequency and intensity of the stimulus have been described as variables that may influence seizure induction.

Interestingly, our patient described a very narrow area of the gums which caused her seizures.

From a pathophysiological and anatomical point of view, we believe that the stimulus is driven by the maxillary nerve, the branch of the trigeminal nerve which, after its passage to the brain stem, reaches the ventral posteromedial nucleus of the thalamus and projects towards the primary somatosensory cortex in the parietal lobe.

Under normal conditions, brushing of the teeth is perceived as a sensation in the stimulated area. If pain occurs, a reaction will clearly take place away from the nociceptive stimulus. This "withdrawal" behaviour is a motor response that originates in the motor cortex. Abnormal neural connectivity in local cortical dysplasia could "amplify" this response due to electrical dissemination to adjacent motor areas, causing a greater motor response that manifests as a seizure.

Intracranial records of EEGs in focal cortical dysplasia show that the epileptogenic zone may be greater than the lesion identified on MRI, and may originate as complex patterns of propagation to neighbouring or distant regions (Duchowny *et al*., 2000). The precise resection of this "generator" of abnormal electrical activity can explain the excellent clinical response that our patient experienced.

When considering surgery for lesions located within the sensory and/or motor area of the face, the fact that there are bilateral projection pathways of motor and trigeminal sensory information, from and to the cortex, and that these have been identified in humans should be taken into account (Penfield and Boldrey, 1937; Chen *et al*., 1999; Müri, 2016). Initial bilateral cortical responses of somatosensory-evoked potentials with a short latency, in both hemispheres, have been reported following lip tactile stimulation, with no significant differences in their latencies, according to some studies. Bilateral sensation of the tongue following unilateral direct cortical stimulation has been also described in humans (Shibukawa, 2012). Thus, unilateral somatosensory cortex of the facial area could be resected without serious facial sensory deficits, or long-lasting motor deficits, as previous experience has demonstrated (LeRoux *et al*., 1991; Lehman *et al*., 1994).

Conclusion

We present a challenging case of a young woman with severe, disabling, refractory epilepsy, with a predominant reflex component while tooth-brushing or eating. Due to negative MRI studies and being diagnosed with psychogenic seizures, the patient suffered her condition for 23 years. Consideration of reflex epilepsy must be taken into account when facing a case like this. Detailed evaluation within a multidisciplinary team allowed us to consider a possible surgical solution. SEEG helped us to delineate the primary epileptogenic area and eloquent areas, and to perform surgical resection. Motor deficits in the face and tongue were transient, and total control of epilepsy was achieved. □

Legend for video sequence

Case study of a patient with refractory epilepsy, with spontaneous tooth-brushing-induced seizures. The usual seizure sequence consisted of a left hemifacial tingling sensation located around the lips, immediately followed by a left hemifacial tonic contraction, and then by flexion and tonic extension of the left arm, finalizing with clonic contraction of the neck muscles. Anatomical relationships between the lesion and the brain and SEEG implantation scheme are shown. An induced seizure under SEEG monitoring, and motor and sensory stimulation through depth electrodes are presented. Surgical resection and postsurgical neurological examinations are shown.

Key words for video research on www.epilepticdisorders.com

Phenomenology: tooth-brushing-induced seizures, sensory and motor seizure (simple), focal seizure, reflex epilepsy.
Localisation: primary motor and sensory facial areas (right)
Syndrome: reflex epilepsy
Aetiology: focal cortical dysplasia (type II)

Supplementary data.
Summary didactic slides are available on the www.epilepticdisorders.com website.

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None of the authors have any conflict of interest to declare.

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TEST YOURSELF

(1) Neurological deficits associated with the resection of lesions within the facial motor area are usually transient. Explain why.

(2) In the semiological sequence of this patient's seizures, left hemifacial contraction is immediately followed by extension of the arm. Explain anatomically the reason for this.

(3) What factors, pertaining to gingival stimulation, could induce reflex seizures in this case?

Note: Reading the manuscript provides an answer to all questions. Correct answers may be accessed on the website, www.epilepticdisorders.com, under the section "The EpiCentre".