CASE REPORT

Tooth-brushing epilepsy with ictal orgasms

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We report a 41-year-old woman with complex reflex epilepsy in which seizures were induced exclusively by the act of tooth brushing. All the attacks occurred with a specific sensation of sexual arousal and orgasm-like euphoria that were followed by a period of impairment of consciousness. Ictal EEG demonstrated two events of epileptic seizure that were provoked after tooth brushing for 38 and 14 seconds, respectively. The interictal EEG showed epileptiform discharges over the right anterior temporal region and interictal single photon emission computed tomography (SPECT) scan showed relative hypoperfusion in the uncus of right temporal lobe. Brain magnetic resonance imaging (MRI) revealed right hippocampal atrophy. We suggest that tooth-brushing epilepsy, especially with sexual ictal manifestations, may provide insight into the cerebral pathophysiology at the right temporolimbic structure.

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Key words: tooth-brushing epilepsy; orgasm; reflex epilepsy; right temporolimbic structure.

INTRODUCTION

Reflex epilepsy comprises a group of syndromes characterised by seizures that reliably occur in response to a specific stimulus, either simple or complex¹. Tooth-brushing epilepsy is an unusual form of complex reflex epilepsy in which seizures are precipitated exclusively by the act of tooth brushing²⁻⁴. It is usually reported to begin with a partial-onset motor seizure that may be followed by a generalised convulsion²⁻⁴. The semiology suggests that tooth brushing-induced seizures are usually extratemporal in origin. Sexual manifestations such as genital sensations or orgasms have been well documented in the patients with epilepsy, especially in women with temporal lobe epilepsy^{5–7}. However, these manifestations as an ictal component in reflex epilepsy have been infrequently reported. In this article we report on a 41-year-old woman with tooth-brushing epilepsy in whom orgasm-like euphoria is presented as an ictal manifestation.

CASE REPORT

A 41-year-old right-handed woman was diagnosed with intractable epilepsy and referred to our epilepsy clinic. At the age of 22, she had received an abdominal total hysterectomy due to uterine leiomyomas with bleeding. She had not had any children. After the surgery, she ceased having orgasms during sexual intercourse or masturbation. Two years after surgery, while brushing her teeth, she suddenly felt sexually aroused and experienced orgasm-like euphoria very similar to orgasms during coitus. The erotic feeling was followed by a period of impairment of consciousness lasting for about 2 minutes. There were no jerky movements or convulsions. For the next 5 years, she experienced recurrent episodes of these 'unknown' orgasms approximately twice every week and occurring exclusively during tooth brushing. She believed that she was possessed by a demon and felt shame and fear. Nevertheless, after three injuries due to the episodes of impairment of consciousness she started to seek

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medical advice at the age of 29. She was diagnosed with complex partial seizures and carbamazepine therapy was started. However, her seizures could not be controlled by carbamazepine (600 mg/day) alone and valproate (1500 mg/day) was added.

Tracing back her seizure history, all seizures occurred during the complete act of tooth brushing (using toothbrush and toothpaste together). Apart from the act of tooth brushing, no other trigger factor could be identified. Prolonged stimuli to the teeth and gums by chopsticks or fingers did not provoke the seizures. The seizures could not be induced by either the smell of toothpaste or the shaking movement of right hand. She did not think that brushing a specific side or area of her mouth or a specific brand of toothpaste or toothbrush was particularly associated with the occurrence of the seizures. She has never had spontaneous seizures.

The patient's gestational and neonatal history was normal. There was no family history of epilepsy. Physical and neurological examinations, and routine blood tests were normal. Interictal EEG showed spikes in the right anterior temporal area. There was no photosensitivity. Under EEG monitoring, two events of epileptic seizure have been provoked after tooth brushing for 38 and 14 seconds, respectively (Fig. 1). An interictal single photon emission computed tomography (SPECT) scan showed a relatively decreased uptake of radioactivity in the uncus of right temporal lobe. Brain magnetic resonance imaging (MRI) revealed right hippocampal atrophy (Fig. 2).

Although the combination of carbamazepine (600 mg/day) and valproate (1500 mg/day) decreased the seizure frequency to approximately 2–3 times monthly, prolonged (>2–3 minutes) and vigorous

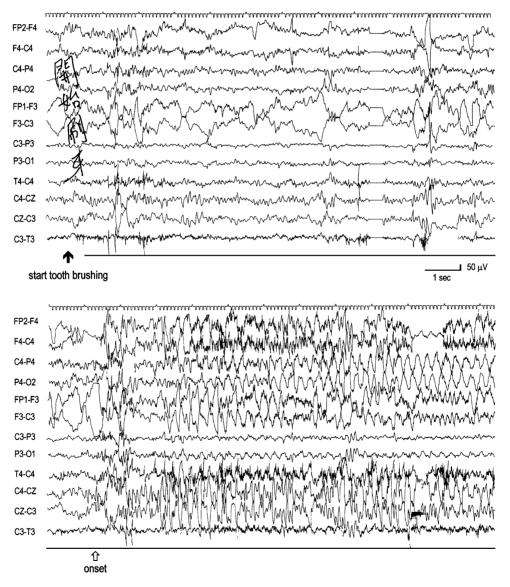


Fig. 1: Ictal EEG of tooth-brushing seizure. The first arrow (♠) indicates a start of tooth brushing. The second arrow (♠) corresponds to occurrence of orgasm and impairment of consciousness after tooth brushing for 14 seconds.

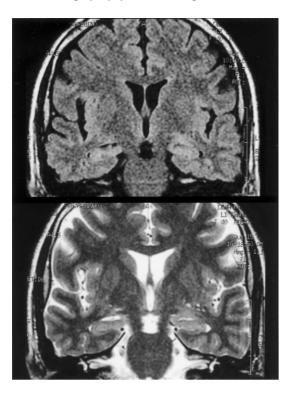


Fig. 2: Brain MRI, coronal view of T1- and T2-weighted images, reveals right hippocampal atrophy.

brushing still could provoke seizures. Consequently, she always shortened the duration of tooth brushing or used mouthwash to care for her oral hygiene. We did not adjust the antiepileptic drugs any more.

DISCUSSION

Our patient demonstrates a very unusual form of complex reflex epilepsy with seizures precipitated exclusively by tooth brushing. No spontaneous seizures occurred in this patient. To our knowledge, only limited cases of tooth brushing-induced epileptic seizures have been reported²⁻⁴. Seizures in these cases were usually simple or complex partial attacks that characterised by an extratemporal origin and may be followed by a generalised convulsion²⁻⁴. Two reported cases had normal neuroimaging results, but EEG revealed left frontal epileptiform discharges^{2,3}. The brain MRI of another case reported by O'Brien et al.⁴ demonstrated a right posterior frontal tumor predominantly involving the precental gyrus with ictal EEG and SPECT scan showing right posterior frontal onset. Our patient had a specific sexual sensation of orgasm during tooth brushing-induced complex partial seizures. The semiology was suggestive of her seizures being mainly involved in the temporolimbic structure.

Sexual sensations or behaviour as a direct manifestation of seizures are rare and have been well documented in patients with temporal lobe epilepsy, especially in women⁷. The preponderance of women with sexual seizures may imply a different neural organisation between male and female sexual functions in the human brain⁷. Epilepsy and sexual activity may be related in some ways. Seizures may be triggered by sexual activity such as hyperventilation provoked during sexual intercourse and reflex epilepsy provoked by sexual stimuli⁵. Furthermore, genital manipulations may occur as a part of an automatism^{5,7}. In our patient, orgasms occurred before the complex partial seizures. However, using an EEG monitoring observation, we excluded the possibility of sex-induced seizures and sexual automatisms in this patient.

The right hemisphere plays an important role in human sexual functions. The production of hormones controlling sexual functions shows a right-side predominance⁸. Sexual arousal is associated with right hemispheric activation. During masturbation-induced orgasms in men, hyperperfusion in right hemisphere has been detected⁹. Sexual ictal manifestations in patients with epilepsy have a lateralising value in the determination of dominant–nondominant relations¹⁰. Orgasmic aura is suggested to be an ictal lateralising sign for the right hemisphere¹⁰.

Different from all those reported cases whose epileptic foci mainly located in the frontal area, the interictal EEG of our patient showed epileptiform discharges over the right anterior temporal region. The findings of the interictal SPECT scan and brain MRI further supported the notion of right temporolimbic involvement in our patient. Therefore, we hypothesised that tooth brushing, a highly specific somatosensory stimulus, can cause the hyperexcitability of a small abnormal area of sensorimotor cortex and subsequent driving of the right temporolimbic region through a direct transcortical pathway.

In conclusion, tooth-brushing epilepsy, particularly with sexual ictal manifestations, may provide insight into cerebral pathophysiology at the right temporolimbic structure. Because epileptic sexual sensation is subjective and intimate, it is difficult to evaluate under laboratory tests. Clinical physicians should acknowledge sexual ictal manifestations in reflex epilepsy that may have significant relevance in neuroanatomic abnormality, especially in women.

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