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Tooth-brushing epilepsy: A case report and literature review

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1. Introduction

Tooth-brushing epilepsy is a type of reflex epilepsy, in which seizures are triggered by specific stereotypical stimuli. Seizure control can be achieved with anticonvulsants such as Carbamazepine or Valproate in some patients. When seizures are intractable, surgical excision with electrocorticography (ECoG) can achieve good outcome. Under awake craniotomy, lesions at the rolandic area can be safely resected in some cases.

2. Case

A 31-year-old gentleman had twitching and impaired consciousness triggered by brushing his teeth for five years. He had good past health and family history was unremarkable. The semiology was that after brushing his teeth for more than 15 s, he developed right face and right arm numbness, which progressed to twitching which lasted about 15 s. He then lost consciousness and developed generalized tonic-clonic seizures (GTC), which subsided spontaneously within one minute. It was noted that his GTC were triggered whenever his teeth were brushed, such as by either hand, by another person, or with an electric toothbrush. As a result, he gave up brushing all together in order to avoid triggering GTC. The impression was complex partial seizures (CPS) with secondary generalization. Physical examination revealed poor dental hygiene (Fig. 1A). There was no focal neurological deficit. Routine blood tests were normal. Computed tomography (CT) of the head was unremarkable. Magnetic resonance imaging (MRI) of the head with contrast showed a 1 cm T2 hyperintense lesion at the pre-central gyrus of the left frontal lobe over the inferior motor strip (Fig. 2C and D), which was consistent with the orofacial motor cortex. Differential diagnoses included a cyst with proteinaceous content or a low-grade glioma. Interictal electroencephalography (EEG) showed rhythmic theta discharges at the left fronto-central region, consistent with the area of the left pre-central gyrus lesion. No ictal event could be precipitated during this investigation. Functional MRI during tasks such as speech, arm movement or tongue protrusion showed no activity at the lesion.

There were three episodes of GTC in two months before treatment. On Valproate sodium 500 mg twice a day he had no further episodes of CPS or GTC but his right face and arm numbness persisted whenever he attempted to brush his teeth. Plasma Valproate level was 422 μmol/L which was within therapeutic range. However, he wanted to stop Valproate due to the side effect of somnolence that had led him to terminate his occupation as an air-conditioner technician. The dosage of Valproate sodium was

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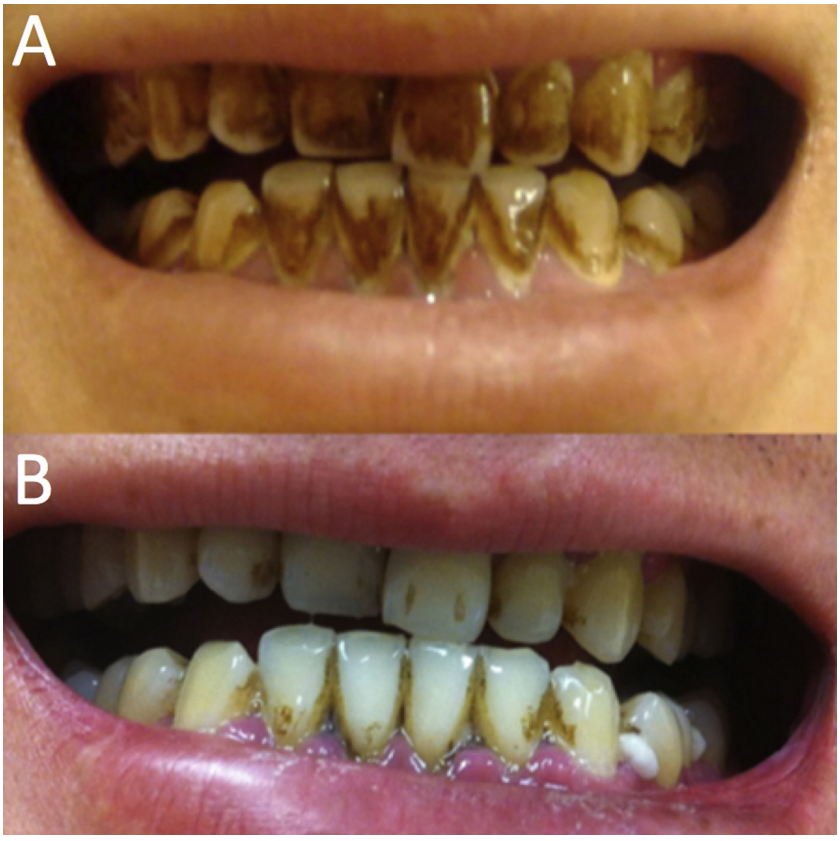

Fig. 1. Clinical Images—A: Preoperative clinical photo showing poor dental hygiene. B: Postoperative clinical photo showing improved dental hygiene.

titrated down to 400 mg twice daily which led to a sub-therapeutic level of Valproate at 64 μmol/L and the re-occurrence of GTC at a frequency of once a month. The anticonvulsant was switched to Levetiracetam 500 mg twice a day and the frequency of GTC was worse at three times in one month. With increasing Levetiracetam dosage to 1000 mg twice a day his GTC frequency improved to once every two months. Clobazam 5 mg daily was also added without further improvement. With suboptimal control by pharmacological treatment, surgical excision was considered. A favorable surgical outcome was anticipated as the lesion found on MRI was concordant with the semiology and EEG findings. The patient refused further titration of anticonvulsants and strongly requested for surgery. Also the nature of the lesion remained uncertain without histology. In view of this, surgical excision of the lesion was performed with electrocorticography (ECoG) (Fig. 3G and H). Intraoperative navigation and awake cortical mapping revealed a firm nodular lesion at the subcortical region of the orofacial motor cortex (Fig. 3I). Excision of the radiological lesion alone was shown intraoperatively to be inadequate. The epileptiform discharges (sharp waves) from the intraoperative ECoG only disappeared when the *peri*-lesional adjacent tissue over the somatosensory area was also excised and thus an extended excision of the lesion was performed. Histology showed gangliocytoma (Fig. 3J).

Postoperatively, there was transient tongue weakness with deviation towards the right, which resolved spontaneously. He made good recovery and was discharged on postoperative day 3. He started brushing his teeth on postoperative day 5 with no aura or seizures. The patient decided to discontinue anticonvulsants one month after the operation. He tolerated tooth-brushing well. Follow-up MRI one year after the operation showed complete excision with no recurrence (Fig. 2E and F). He had remained seizure free for two years after the excision and had not been on any anticonvulsant. He had no neurological deficit. Without seizure, he was able to brush enabling better oral hygiene (Fig. 2B).

3. Discussion

Tooth-brushing epilepsy is a rare type of reflex epilepsy or stimulus-evoked epilepsy. To the best of our knowledge, there were sixteen reported cases of tooth-brushing epilepsy in the literature [1], [2], [3], [4], [5], [6], [7], [8], [9], [10], [11], (Table 1). It was first described in 1982 in a twelve-year-old boy with stereotypical Jacksonian "marching" seizures evoked by tooth-brushing stimulation [1]. An association with a structural lesion was first demonstrated in 1996 with MRI, EEG and single-photon emission computed tomography (SPECT) showing a corresponding frontal lobe lesion [2]. However, EEG could be the only positive investigation and no structure abnormality identified in MRI [4], [7], [9], [11]. As in our patient, epilepsy could also be induced by an electric toothbrush, as first described in 2008 [4], or passive tooth-brushing, as reported in 2013 [10]. In contrast to our patient, there could be no motor symptoms but only sensory or autonomic symptoms followed by loss of consciousness [4], [8].

Epilepsy induced by somatosensory stimulus is uncommon. Examples of possible triggers include patting on the shoulder, rubbing of the leg or scratching of a confined area of scalp. These were coined "rub epilepsy" in 2001 [5]. In those patients, the MRI were normal and the semiology started with a "sensory Jacksonian march". It was previously suggested that tooth-brushing epilepsy was a separate entity in which it usually demonstrated a "motor Jacksonian march" with no sensory symptoms[5]. Our case showed both "sensory and motor Jacksonian march" with the semiology of right arm and right face numbness followed by twitching. Other cases of tooth-brushing epilepsy in the literature also show both entities (Table 1). In our patient, seizures were also elicited by

Table 1
Summary of reported cases of tooth-brushing epilepsy in the literature.

Case	Authors	Year	Age/Gender	Other triggers	Onset symptoms	Seizures	Symptoms	MRI	EEG	Functional scan	Medication	Surgery/Pathology	Treatment response
1	[1]	1982	12/M	Maxillary palatal expansion device	"R face tingling and spasms"	SPS	S, M	CT normal. No MRI.	L fronto-central	–	PHT, CBZ	–	No seizures
2	[2]	1996	9/F	Startle/anxiety	"Numb feeling in head"	SPS, CPS	S, M	R *peri*-rolandic region	R fronto-central	Ictal SPECT – R frontal hyperperfusion	PHT, VPA, TPM, CBZ	–	No seizures
3–7	[3] (5)	1996	–	Startle (1) Eating or drinking (2)	Twitching of R corner of mouth (5)	SGS (2)	M (5)	Atrophy (2) Cortical dysplasia (2) Gliosis (1)	L central temporal (4)	–	–	Yes (4)/pathology not reported	Less frequent twitching with no GTC (2)
8	[7]	2001	28/F	None identified	"Mouth and gum numbness"	CPS, SGS	S, M	Normal	L superior frontal	–	CBZ	–	No seizures
9	[5]	2001	3/F	Eating solid food	"Twitching of L corner of mouth"	SPS	M	Not reported	R central	–	–	–	–
10	[8]	2004	24/F	None identified	"Orgasm-like euphoria"	CPS, SGS	S, A	R hippocampal atrophy	R anterior temporal	Ictal SPECT-R temporal hypoperfusion	CBZ, VPA	–	Seizures 2–3 times monthly with tooth-brushing
11	[9]	2006	50/M	Thoughts of toothbrush	"Pleasant feeling"	CPS, SGS	M	Normal	L temporal	–	CBZ, TPM, LTG, CLN, PHB, VPA, OXC, LEV, PGB	–	One seizure per week
12–13	[6] (2)	2007	27/M	Eating potato chips	"Tightening/tingling in tongue"	SPS, SGS	S, M, A	L post-central gyrus	L parasagittal and temporal	FDG PET-Normal	CBZ	–	Monthly SPS with tooth-brushing
			36/M	Eating	"Cramping sensation in tongue and jaw"	SPS, CPS, SGS	S, M	R post-central gyrus	Possibly R postictal temporal	FDG PET-Normal	–	2 resections/Cortical dysplasia	No seizures
14	[4]	2008	11/F	Powered toothbrush	Abdominal discomfort	CPS, SGS	S, A	Normal	L frontal central temporal	–	CBZ, VGB, VPA, OXC	–	No seizures
15	[10]	2013	8/F	Passive tooth-brushing	Not reported	CPS, SGS	M	Slight lateral ventricles enlargement	Bilateral frontal	Ictal SPECT-R BG & L *peri*-sylvian hyperperfusion	PHT, VPA, CBZ, ZNS, CLB, SUL, TPM, CLX	–	Seizures several times a day
16	[11]	2014	47/M	None identified	Eyes wide opened	SPS, CPS	S, M, A	Normal	L central	–	LEV, LAC	–	No seizures
17	Current case report	2016	26/M	Passive tooth-brushing, powered toothbrush	R arm & face numbness	SPS, CPS, SGS	S, M	L pre-central gyrus	L fronto-central	fMRI (speech, tongue and hand movement), DTI	VPA, LEV, CLB	Awake cran-iotomy/gangliocytoma	No seizures

Abbreviations- (): number of patients; R: right; L: left; –: not performed/not available. Seizures— SPS: simple partial seizures; CPS: complex partial seizures; SGS: secondary generalized seizures. Symptoms— S: Somatosensory symptoms; M: Motor symptoms; A: Autonomic symptoms. Imagings— CT: computed tomography; MRI: magnetic resonance imaging; fMRI: functional magnetic fesonance imaging; DTI: diffusion tensor imaging; SPECT: single-photon emission computed tomography; FDG PET: F-18 fluorodeoxyglucose positron emission tomography; BG: basal ganglia. Medications—PHT: phenytoin; CBZ: carbamazepine; VPA: valproate; TPM: topiramate; LTG: lamotrigine; CLN: clonazepam; OXC: oxacarbazepine; LEV: levetiracetam; CLB: clobazem; LAC: lacosamide; PHB: phenobarbital; PGB: pregabalin; VGB: vigabatrin; ZNS: zonisamide; SUL: sulthium; CLX: cloxazolam.

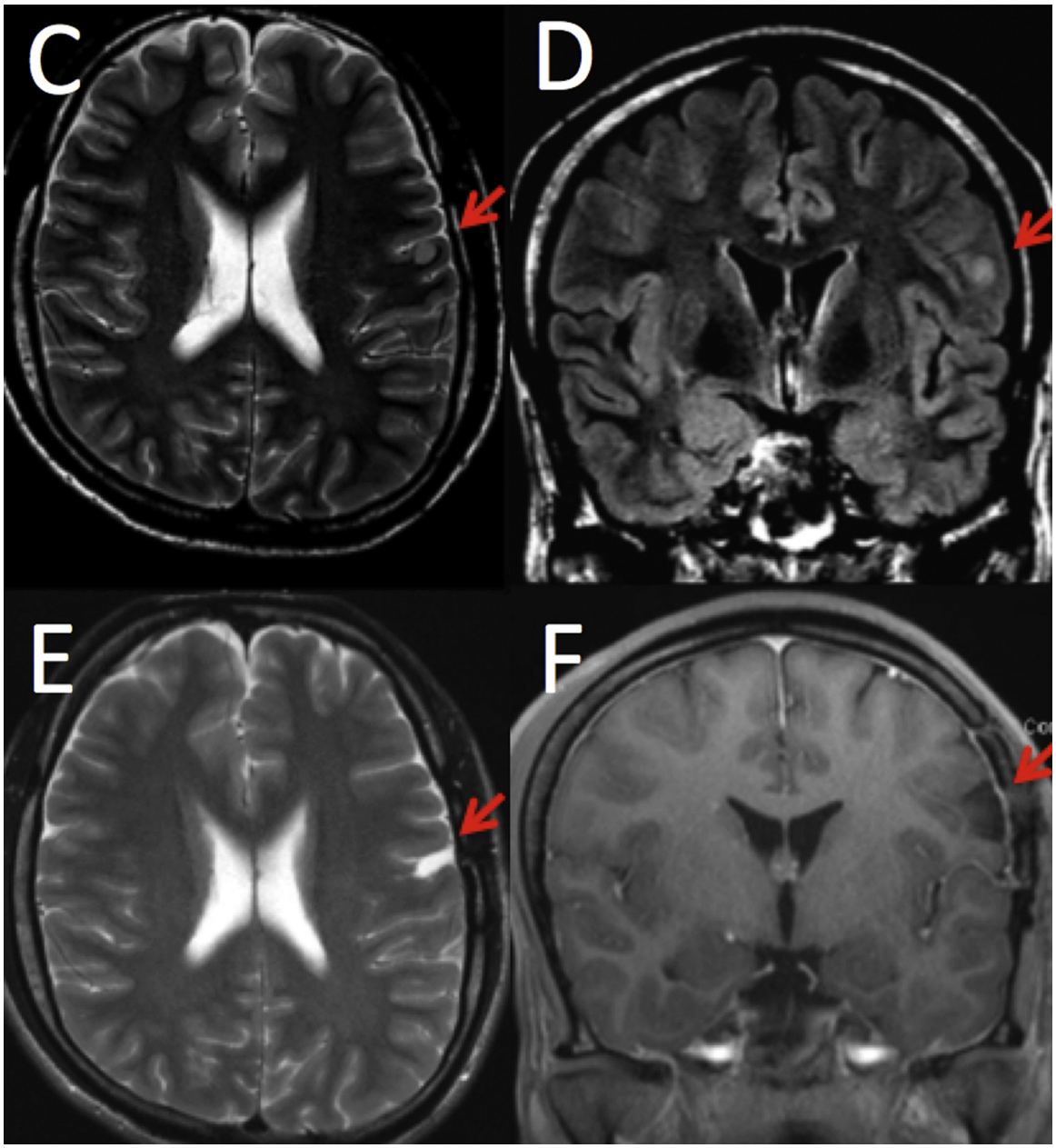

Fig. 2. Radiological Images—C: Preoperative MRI axial T2 showing 1 cm hyperintense lesion at the pre-central gyrus of the left frontal lobe (red arrow indicates lesion). D: Preoperative MRI coronal T1 contrast showing 1 cm hyperintense lesion at the inferior motor strip (red arrow indicates lesion). E: Postoperative MRI axial T2 showing complete excision of the lesion with no recurrence (red arrow indicates location of previous lesion). F: Postoperative MRI coronal T1 contrast showing complete excision of the lesion with no recurrence (red arrow indicates location of previous lesion) (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

other people brushing his teeth and using an electric toothbrush. This implies that the trigger was independent of his own rhythmic brushing motion. Hence, it was highly suggestive that the seizure onset zone was located in the sensory area, in which a somatosensory stimulation provoked the propagation of epileptic discharges.

In general, Carbamazepine is the most logical choice for focal epilepsy. It can be prescribed alone or combined with Valproate [12], [13], [14]. For the cases of tooth-brushing epilepsy reported in the literature, more than half became seizure-free on Carbamazepine or Valproate [1], [2], [7], [4], [11], (Table 1). However, there were significantly more patients on Carbamazepine developing skin rash as a side effect [15]. Previous studies conducted in Hong Kong and Taiwan found 100% association between allele HLA-B*1502 and Carbamazepine-induced Stevens-Johnson syndrome (SJS) in Han Chinese in Southeast Asia [16], [17], [18]. At the same time, some patients with Carbamazepine-induced SJS were negative for HLA-B*1502, but they carried some other associated HLA-B alleles [19]. Hence, the risks of developing severe cutaneous reactions in Han Chinese patients receiving Carbamazepine remained regardless of the HLA-B*1502 status. As a result, our institution did not prescribe Carbamazepine as a first line treatment but instead prescribed Valproate. In our patient this led to good seizure control but the side effects were not well tolerated.

Similarly to our patient, there had been cases of tooth-brushing epilepsy where the lesions were located at the *peri*-rolandic region [2], [6]. In our case, an extended excision of the radiological lesion had to be performed as the *peri*-lesional tissue was epileptogenic. This demonstrated that the epileptogenic zone (region of which

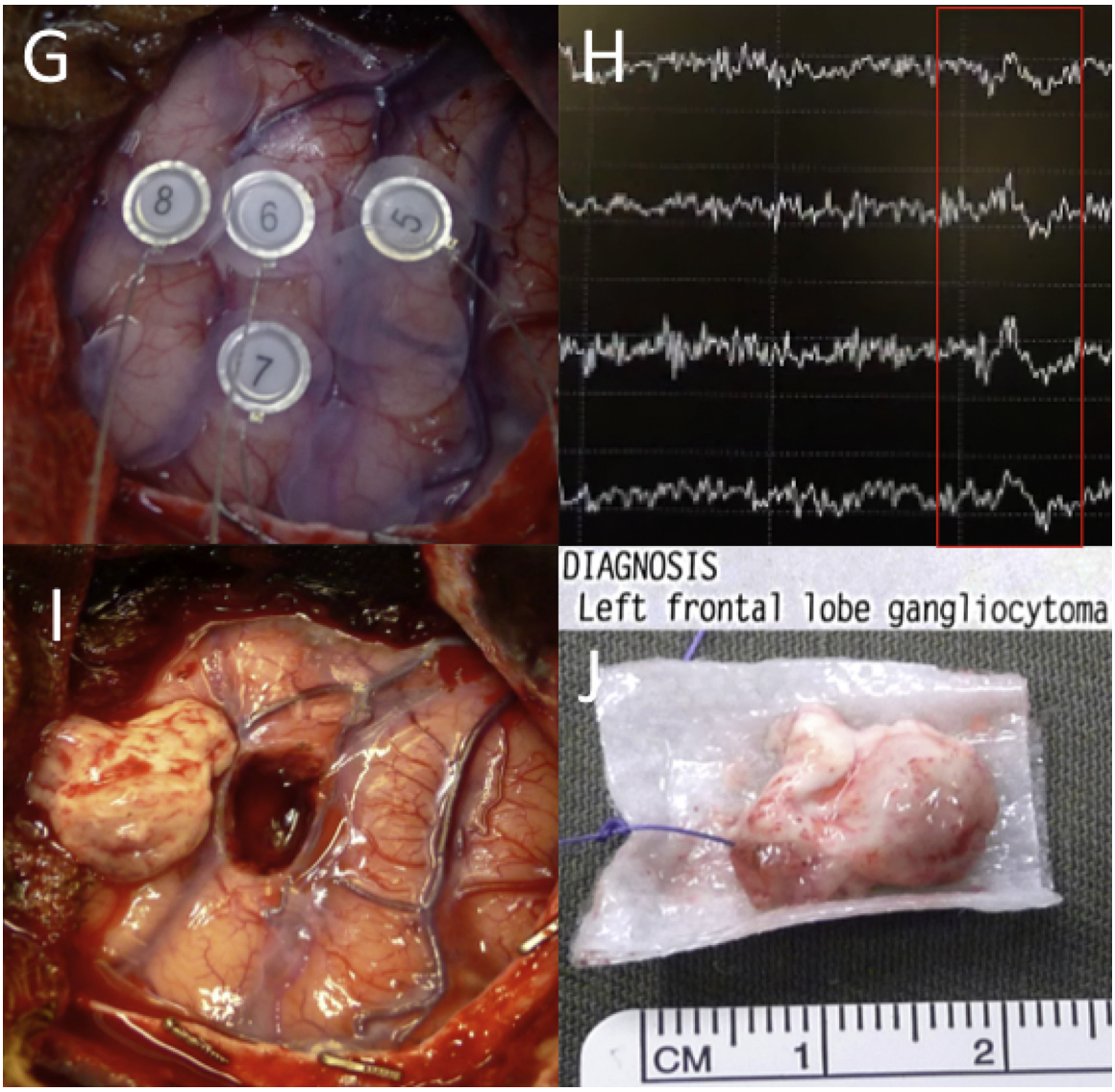

Fig. 3. Intraoperative Images—G: Intraoperative electrocorticography electrodes placement. The central sulcus was located between electrode "6" and "8". Awake cortical mapping demonstrated that area of "6" was oral motor cortex, "7" was the facial motor cortex and "8" was facial somatosensory cortex. "5" was not an eloquent area during mapping. H: Intraoperative electrocorticography (the red box showing spikes and sharps, which were compatible with the identification of the epileptogenic zone). I: Intraoperative image showing excision of the lesion together with the epileptogenic *peri*-lesional tissues and the overlying cortex. J: 1 cm lesion with about 0.5 cm *peri*-lesion tissues and overlying cortex. Pathology reported as gangliocytoma.

resection was necessary and sufficient to achieve seizure freedom), which extended to the somatosensory cortex, was larger than the epileptogenic lesion (structural lesion which was causally associated with epilepsy), which was located at the motor cortex. Thus intraoperative ECoG was an invaluable tool when excising such lesions. It was likely that the irritation zones (regions that were not epileptogenic themselves but may produce interictal spikes) were at the sensory motor cortex, whereas the seizure onset zone (region initially generating ictal discharges) and the symptomatogenic zone (region that produced the first clinical symptoms) were at the somatosensory cortex [20].

We also demonstrated that resections over the rolandic cortex could be safely performed even over the orofacial motor cortex of the dominant hemisphere. With proper preoperative planning, intraoperative awake cortical mapping and electrocorticography, good outcome can be achieved.

4. Conclusion

Tooth-brushing epilepsy remained an interesting challenge for neurologists and neurosurgeons. Seizure control can be achieved with anticonvulsants such as Carbamazepine or in combination with Valproate in some cases. When seizures are intractable, we have shown that surgical excision with intraoperative ECoG can achieve good seizure-free outcome. Under awake craniotomy, lesions at the rolandic area can be safely resected in some cases.

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