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Lingual myoclonus associated with brain tumour: an epileptic origin?

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ABSTRACT – We describe a 59-year-old man, suffering from a left-sided weakness, who was diagnosed with a right frontal oligodendroglioma. One month after a craniotomy, he complained of perioral numbness and slurred speech, which lasted about an hour. Neurological examination discovered dysarthric speech and repetitive, rhythmic myoclonic jerks of the tongue and the lower lip. We suggest this lingual myoclonus was an epileptic phenomenon associated with brain tumour. *[Published with video sequences]*

Key words: myoclonus, lingual, epileptic, brain tumour, epilepsia partialis continua

Isolated lingual myoclonus is an uncommon and poorly understood clinical phenomenon (Sridharan, 1984; Bettoni *et al.*, 1999). It usually is observed with palatopharyngeal myoclonus and associated with brainstem pathologies (Kulisevsky *et al.*, 1994; Postert *et al.*, 1997). Li *et al.* (2010) described a case of lingual myoclonus associated with cerebral cavernoma, and suggested an epileptic origin. We report a patient who presented with lingual myoclonic jerks and cerebral glial tumour.

Case study

A 59-year-old man, suffering from a left-sided weakness, was diagnosed with a brain tumour on the right frontoparietal lobe. He was operated on and the pathological diagnosis was oligodendroglioma. Phenytoin (200 mg/day) was added, for the prophylaxis of eventual postoperative seizures, to his amlodipine treatment for essential hypertension. One month after the operation, he was hospitalized for the rehabilitation of the left hemiparesis. His initial neurological examination also disclosed a left facial paresis of central type, hyperreflexia, and left-sided hemisensory deficit. The psychiatric examination revealed a major depression and anxiety disorder, and a medical treatment with sertraline, 100 mg/day and mirtazapine, 30 mg/day, was proposed.

One month after the hospitalization, he complained of perioral numbness and slurred speech, which lasted about an hour. An EEG was recorded only after the jerks disappeared spontaneously after about an hour, and no abnormal epileptiform activity was observed. The same complaints restarted six hours later, and neurological examination discovered dysarthric speech and repetitive, rhythmic jerks of the tongue and the lower lip from right to the left side, with a frequency of 1 jerk/sec, without any accompanying cervical, palatal or pharyngeal abnormal

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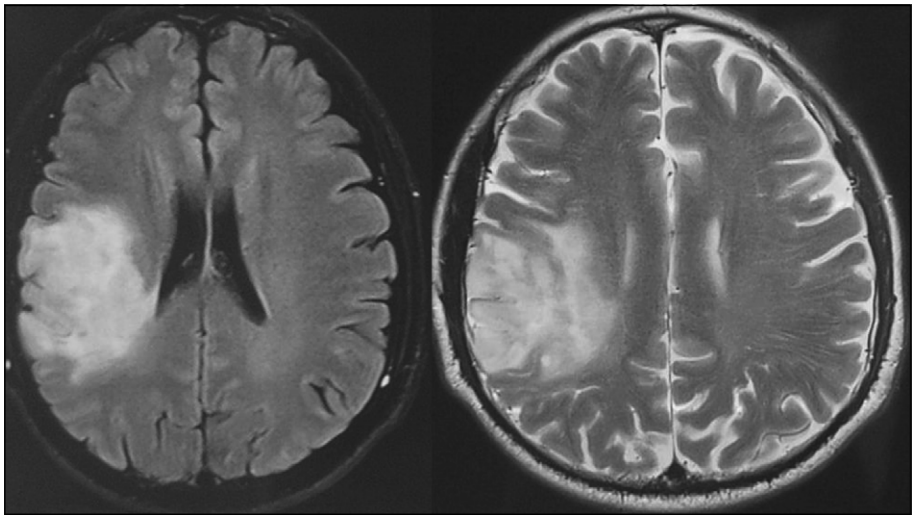

Figure 1. Brain MRI showing a right frontoparietal tumour.

movements *(see video sequence)*. Lingual myoclonic jerks being contralateral to the cerebral lesion, we diagnosed *epilepsia partialis continua*.

We replaced phenytoin with sodium valproate, 1,000 mg/day *per os*, to avoid an eventual enzyme-inducing effects of phenytoin in a patient on multiple drug therapy. A cranial CT scan excluded a cerebral haematoma and MRI showed residual glial tumour *(figure 1)* and intact brain stem *(figure 2)*. Sodium valproate was discontinued two weeks later and lingual jerks did not recur.

Six months later, the patient complained of recurring slurred speech and tongue numbness, with some left-sided facial clonic movements, which recovered spontaneously after about an hour. Neurological examination was the same as before, any abnormal movement was not observed, and a control EEG was also normal.

Discussion

The pathophysiology of lingual myoclonus is not well understood and the epileptic origin not well documented. Jabbari and Coker (1981) reported three children with paroxysmal and rhythmic lingual movements and speculated an unusual form of "subcortical seizures". Li *et al.* (2010) described a case of lingual myoclonus associated with an MRI-documented cerebral cavernoma over the left frontal region, interictal epileptiform EEG abnormality and complete resolution of tongue movements by antiepileptic drug treatment, and suggested an epileptic origin. In our case, we observed contralateral rhythmic jerks of the tongue, from the right to the left side, and demonstrated a cerebral tumour on the right side, which involved the tongue motor area of the motor cortex, as in the case described by Li *et al.* (2010). Therefore, we also suggest an epileptic origin, although the interictal EEG did not show any epileptiform abnormality, presumably due to the very localized myoclonus. In conclusion, isolated lingual myoclonus can be considered as a rare manifestation of epilepsia partialis continua. □

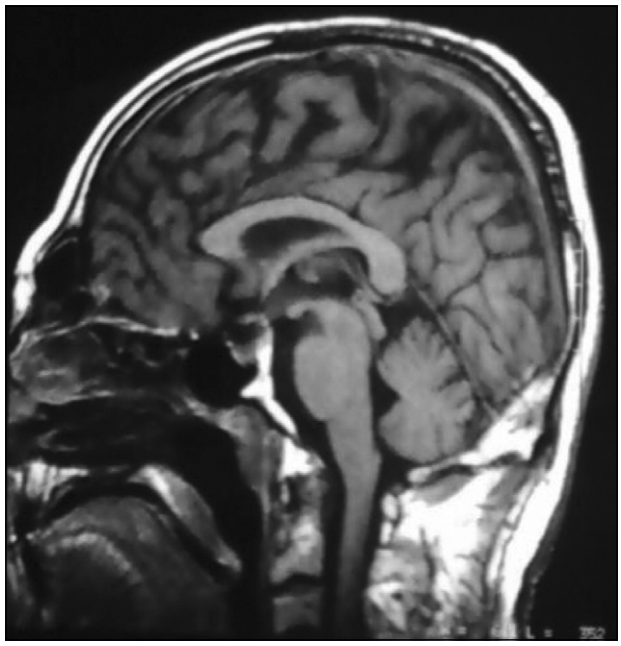

Figure 2. Brain MRI showing intact brain stem.

Legend for video sequence
Rhythmic jerks of the tongue.

Disclosure.
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