Letter to the Editor

Symptomatic palatal tremor following multiple listerial brainstem abscesses

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Palatal tremor is a rare movement disorder characterised by continuous involuntary rhythmic movements of the soft palate, sometimes including other muscles innervated by cranial or spinal nerves. The condition may be essential or symptomatic [1]. We describe a case of symptomatic palatal tremor associated with hypertrophic olivary degeneration (HOD) following multiple brainstem abscesses caused by Listeria monocytogenes. To our best knowledge, this association has not previously been reported in the literature.

A 45 year old man with history of hepatitis C was admitted with a five day history of headache, vomiting, diplopia, left sided facial numbness and left sided limb weakness. Clinical examination demonstrated bilateral abducens nerve palsies, left peripheral facial nerve palsy, sensory impairment in the ophthalmic distribution of left trigeminal nerve and left sided pyramidal weakness. Contrast enhanced T1 weighted magnetic resonance imaging (MRI) of the brain demonstrated multiple ring enhancing lesions in the brainstem suggestive of multiple abscesses. These lesions were hyperintense on diffusion weighted imaging (DWI) with corresponding hypointensity on the apparent diffusion coefficient (ADC) sequences, consistent with restricted diffusion seen in brain abscess (Fig. 1A–D). Blood cultures were positive for L. monocytogenes, presumably of dietary origin. HIV serology was negative and echocardiography was normal. Lumbar puncture was not performed. He was treated with ampicillin and gentamycin for three months. He made a remarkable improvement but had residual oscillopsia and left peripheral facial nerve palsy. He was discharged home and had no difficulties in performing activities of daily living.

At eight months follow up, he reported constant clicking in his ears. Examination demonstrated palatal tremor of approximately 3 Hz (video 1) which persisted in sleep. Repeat brain MRI demonstrated a T2 hyperintense and hypertrophied lesion in the right inferior olivary nucleus consistent with HOD (Fig. 1E&F). The previously seen pontine changes had resolved.

Supplementary video related to this article can be found at http://dx.doi.org/10.1016/j.parkreldis.2013.11.003.

Listeriosis is an uncommon infection caused by L. monocytogenes, a gram positive facultative intracellular anaerobic bacterium that has tropism for meninges and brain parenchyma. CNS manifestations include meningoencephalitis, rhomboencephalitis, cerebritis and abscesses. Basal meningoencephalitis is the most common CNS manifestation while brain abscess is rare, representing less that 10% of all CNS listeriosis [2,3]. Patients with listerial brain abscesses are almost always bacteremic and 38% of the cases have concomitant meningitis, a feature that is unusual in other forms of bacterial brain abscess [2,3]. Cerebrospinal fluid and blood cultures are positive in 38% and 86% of the cases respectively [2,3]. There is a high mortality rate of 40–51% which is higher than in non-listerial brain abscess and approximately 61% of the survivors are left with some neurological deficits [2,3].

Palatal tremor is an uncommon movement disorder and is classified into essential and symptomatic forms. Symptomatic palatal tremor (SPT) results from lesions affecting the dentato-rubro-olivary pathway or the Guillain–Mollaret triangle [1]. This triangle is a functional circuit connecting the dentate nucleus of the cerebellum on one side with the red nucleus and the inferior olivary nucleus on the other side, via the superior cerebellar peduncle, the central tegmental tract and the inferior cerebellar peduncle, respectively. Essential palatal tremor (EPT) has no demonstrable cause and no radiological features of HOD [1]. Patients with EPT may complain of rhythmic objective ear clicks which are rare in the symptomatic form. Patients with SPT may manifest other clinical features of brainstem or cerebellar dysfunction including nystagmus, dysarthria and ataxia [1].

SPT is often associated with HOD, a unique transsynaptic degeneration secondary to injury to the dentato-rubro-olivary pathway [1,4]. MRI may show non-enhancing T1 isointense and T2 hyperintense enlargement of the affected olive. The lesion causing HOD is usually in the contralateral dentate nucleus or ipsilateral brainstem [1,4]. Common insults leading to HOD include haemorrhage, infarction, demyelination and tumours [1,4,5]. Palatal tremor is rarely associated with an infectious process which may include syphilis and encephalitis [1]. No report of palatal tremor subsequent to CNS listeriosis was identified in the literature.

Development of SPT is temporally associated with signal changes and hypertrophy of the olivary region [4,5]. SPT commonly appears within few weeks or months after an insult to the Guillain–Mollaret triangle, reaching a peak between 5 and 24 months [5]. Hyperintense olivary signal on T2 weighted MRI usually develops by 1 month after the inciting lesion and may persist for a number of years or indefinitely; olivary hypertrophy develops around 6 months after the event.
and usually persists for 3–4 years, ultimately leading to shrinkage [4]. Clinical symptoms including palatal tremor usually persist.

In conclusion, development of SPT 8 months after a brainstem infection in our patient is consistent with the temporal evolution of HOD in other conditions, and this report demonstrates that CNS listeriosis can be a cause.

Author contributions

Suresh Kumar Chhetri: study concept and design, drafting/revising the manuscript for content, data interpretation, coordination and execution of the study.

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