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Intermittent undulating tongue as an involuntary movement in early amyotrophic lateral sclerosis

A pillar of movement disorders neurology is the accurate characterization of phenomenology to inform the precise identification of a clinical syndrome, helping to pursue a prioritized and efficient differential diagnosis. We present a case of very rare lingual hyperkinesia named undulating or galloping tongue that literature suggests to be associated with a topographical diagnosis [2,4].

A 51-year-old right-handed previously healthy male presented with a 7-month history of involuntary movements of the tongue. He had slow and strained speech, drooling and dysphagia. The onset of his symptoms was coincident with the occurrence of an upper respiratory infection. When drinking liquids, he would at times hold his mouth closed with his fingers. Otherwise, he had the sensation that his tongue would would protrude and expel liquid out of his mouth. There were no other involuntary movements. He had not noticed any limb weakness or dyspnea. The initial neurological examination documented an undulating motion of the entire tongue along its longer axis that was present more prominently in a semi-protruded position. At rest, the examiner observed a discrete and recurrent protrusion of the tongue. When the patient spoke, ate a cracker or drank water from a cup, there was no protrusion of the tongue. No palatal tremor was observed. There was a mild spastic dysarthria and mild atrophy of the tongue (video 1). The tongue movements were not distractible. There was no other hyperkinesia. Limb muscle power was normal. There was hyperreflexia at the triceps, however other deep tendon reflexes were normal, and there was no clonus or Hoffman sign. There was no limb or axial incoordination. Plantar reflexes were flexor. Gait was normal.

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The initial laboratory investigations were normal and included a brain MRI, copper studies, EMG/NCS and antiganglioside antibodies. However, when reassessed five months later the patient had developed new paresis and widespread signs of motor neuron disease. Repeat NCS/EMG were consistent with clinically probable – laboratory supported ALS, according to the revised El Escorial criteria [1]. The bulbar-onset ALS continued to progress overtime and he passed away from his illness approximately 2 years after symptom onset.

An undulating or galloping tongue is a very rare movement disorders phenomenology. It has been described as a "ripple" morphology seen in a liquid surface after the impact of an object [2]. It is challenging to include this type of movement disorder in known categories of hyperkinesia. For example, dystonic movements are typically patterned and twisting, while in this case, the undulating movement is more akin to an oscillation. Tremor is defined as an involuntary, rhythmic, oscillatory movement of a body part [3]; the observed undulating tongue movement corresponds to the coordinated contraction of consecutive tongue areas and not an oscillatory movement of the whole tongue. The undulating tongue does not have a random pattern that defines chorea. There was no phenomenology associated with tics such as the presence of a premonitory urge.

An undulating tongue has been described in Wilson's disease [2] and, more frequently, with brainstem structural lesions of various etiologies. For the first time, an undulating tongue is associated with ALS and as an initial clinical manifestation. It has been hypothesized that lingual dyskinesia may be related to pontine central tegmental tracts in patients with brainstem ischemia [5]. To some extent, an undulating tongue could represent a compensatory mechanism. A ‘release’

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phenomenon of hypoglossal neurons *via* the loss of inhibitory input contained in 'dying' first-order motor neurons may underlie the undulating tongue [5].

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