

Letter to the editor: A case of functional isolated tongue tremor-like dyskinesia after Covid-19 vaccine

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The presumed side effects of the Covid-19 vaccination are under the scrutiny of the scientific community. Although post-marketing surveillance is still ongoing, Covid-19 vaccination with mRNA technology is recognized as highly effective, with an excellent safety profile. Nevertheless, alongside the well-known vaccination-related side effects (fever, headache, or fatigue), neurological symptoms (paralysis, seizures, tremors) have been reported.¹ Some of these symptoms could be framed as Functional Neurological Disorders (FND),²⁻⁵ neurological symptoms affecting motor or sensory function, that cannot be explained by typical neurological diseases or other medical conditions. Functional movement disorder (FMD) is a common manifestation of FND, presenting with different symptoms of altered movement, including tremor, jerks, dystonia, and gait disorders, or with combinations of phenotypes⁶.

We describe herein a patient who presented functional tremor-like dyskinesia of the tongue, notably a very rare clinical manifestation of FMD if in isolated form, following Covid-19 vaccination.

This young University student, in her early 20s, was vaccinated with the first dose of Moderna-Spikevax vaccine in July 2021. During the following 3 days, besides the commonly reported tiredness and fatigue, she suddenly developed irregular involuntary movements of the tongue, that occasionally tended to come out of the mouth. In the following weeks, these manifestations were replaced by an irregular tongue tremor interspersed by choreiform or writhing lingual movements, with an irregular pattern of intensity and frequency, generally conditioned by tension or agitation state. No dysphagia, dysarthria, xerostomia or dysgeusia were reported. In September 2021, the patient was hospitalized at “Istituto Neurologico Carlo Besta” for further investigations. At admission, general physical and psychic examinations were within the normal range. Neurological examination showed a quite variable and diffuse irregular tremor of the tongue, interspersed by choreic-like/jerky movements, however without tongue impersistence, a sign that consists in the involuntary lingual retraction when asked the patient to protrude the tongue, frequently encountered in patients with Huntington’s Chorea or other genetically-determined choreas; her tongue movements were conditioned by distracting maneuvers (consisting in specifically designed exercises aimed to divert subject’s attention, such as

clapping or concentrating on a math problems) and appeared to be variable during the day and to stop during the night rest. Blood tests, electrocardiogram, electroencephalogram, high-definition brain MRI with focus on brainstem and hypoglossal nerves (Figure, panels A1 and A2) and functional study of the cortico-bulbar tracts by transcranial magnetic stimulation (Figure, panel B) were normal. Considering the negativity of the aforementioned examinations and the positive clinical signs, spinal tap, which is an invasive procedure for the patient, was not performed. From a psychopathological angle, she reported to have suffered from depression, anxiety, and insomnia during the last year. During the 9 days-hospitalization a significant improvement of the lingual dyskinesia, up to recurrent long-lasting pauses in dyskinetic manifestations, has been observed by the neurologists. In the light of the clinical features of the disorder and of the negativity of diagnostic-instrumental set, the multidisciplinary staff concluded for a functional dyskinesia of the tongue, according to the diagnostic criteria for Functional Motor Disorders (FMD), in the context of an adjustment disorder with anxiety and depressed mood. At the following evaluations, persistent although variable lingual symptomatology, more relevant in the evening and in association with stressor situations, was reported by the patient. During the last neurological evaluation (May 2022), a more pronounced restless tremorogenic tongue was evident, with a combination of irregular tremor and jerks, but always with no tongue impersistence nor dysphagia or dysarthria.

The precipitating factors for the development of FND after Covid-19 vaccination in people with biological, social, and/or psychological predisposition, are most likely to be ascribed to expectations, beliefs, heightened bodily attention, arousal, and emotional processing. As reported in many studies, abnormal expectations or beliefs can interact with sensorimotor perceptions to provide the mechanistic equation for FND. Currently only sporadic case reports of FND consequent to vaccination for Covid-19 have been described: a 42-year-old female who developed episodic loss of consciousness; a 28-year-old woman presenting paraparesis and bilateral foot drops; a patient with psychogenic non-epileptic seizures; a patient with persistent dizziness and a loss of sensitivity in the right arm and leg.^{7,8}

In our opinion, there are two levels of dealing with this problem: primary and secondary prevention. First, it would be useful to share vaccine safety data with recipients, to allay their excessive emotional involvement; second, acknowledging FND as a side effect of the vaccine, would prevent FND misdiagnosis, and allow the medical community to promptly recognize the nature of the symptom and to investigate it with correct examination, curtailing any prolongation of the disorder.

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Figure captions.

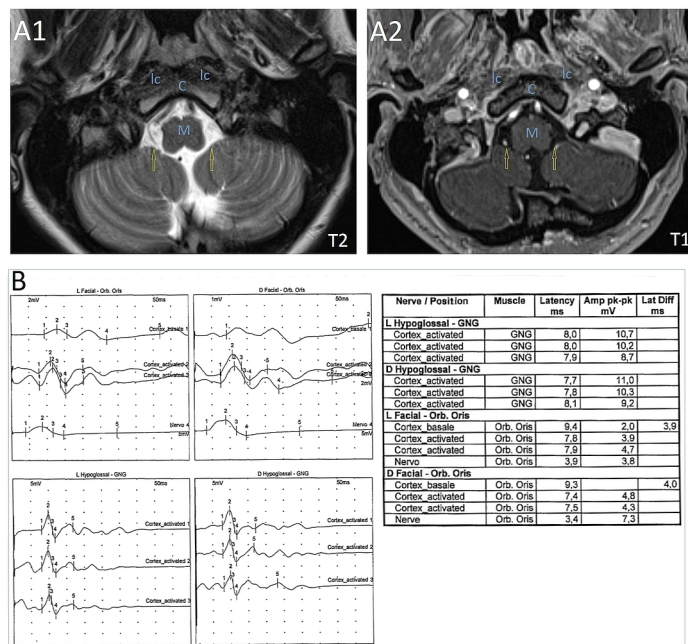
Panel A. MRI study. A1. An Axial fast spin echo T2-weighted image: normal hypoglossal nerves canals (straight yellow arrows).

A2. An axial post-gadolinium T1-weighted image with fat saturation at a similar level, shows normal signal from both hypoglossal nerves canals (straight yellow arrows). No contrast enhancement is present. (M, medulla; C, clivus; lc, longus colli muscles).

Panel B: Functional study of the cortico-bulbar tracts by transcranial magnetic stimulation, showing a normal central motor conduction without side asymmetry.

Supporting Video captions.

Supporting Video: Time-evolution of the clinical features presented by the patient. Three periods are showed, with tongue dyskinesias as on July 2021 - some weeks after onset, on October 2021 - 3 months after onset and on May 2022 - 10 months after onset – at the last FU.



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