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Letter to the Editor: New Observation

Meige Syndrome Following COVID Infection

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A 61-year-old woman known for prediabetes and dyslipidemia was referred by her optometrist to our neurology clinic for blepharospasm. Her past neurological history was unremarkable, and she was not previously exposed to dopamine-depleting agents. She had received three prior doses of COVID vaccination. Her symptoms began 7 days after the first episode of an asymptomatic COVID infection confirmed by molecular testing. This evolved over a period of 2 to 3 months into functional blindness from blepharospasm and speech difficulties due to oral and facial dystonia (video 1, segment 1). There initially was no implication of cervical or axial musculature, but 5 months after the initial presentation, the patient also developed mandibular, cervical, and vocal cord dystonia (video 1, segment 2). This constellation of symptoms led to social isolation, depressive symptoms, and professional disability. A thorough workup was performed, including complete blood count and differential, serum protein electrophoresis, electrolytes, renal function, hepatic function, vitamin B_{12}, C-reactive protein, TSH, anti-TPO, anti-B2-glycoprotein, IgA, anti-gliadin, anti-Sm, anti-RNP, anti-SSA, anti-SSB, and anti-scl 70, and Tropheryma whipplei polymerase chain reaction, which were negative. A normal magnetic resonance imaging excluded structural or vascular lesions. Botulinum therapy injections were initiated with partial success after four cycles. As an adjunctive treatment, a trial of clonazepam produced suboptimal benefit, which was followed by another trial of zolpidem that was later stopped by the patient for lack of benefit. Tetrabenazine was contraindicated due to severe depressive symptoms, and Trihexyphenidyl was refused by the patient due to its side-effect profile.

Meige syndrome is a rare movement disorder characterized by blepharospasm and oromandibular dystonia.[1,2] This focal dystonia typically presents in middle-aged women and is usually idiopathic. Secondary forms exist following dopamine-blocker use or with structural lesions such as strokes, tumors, or demyelination of various brain regions such as the brain stem,[1,3] and the thalamus.[1,3] Some authors report anomalies involving the basal ganglia, the cerebellum, and the midbrain.[3] This syndrome has also been described as a component of other movement disorders such as Parkinson's disease,[1,2] Huntington's disease,[1,2] olivopontocerebellar atrophy,[1] or Lewy body disease.[1] Botulinum toxin injections are the mainstay of treatment, but other reported treatments have included benzodiazepines, anticholinergics, dopamine antagonists, GABA receptor agonists, and zolpidem. Recent studies have also shown the benefit of deep brain stimulation in refractory cases.[1,2] The pathophysiology of Meige has not yet been elucidated, but it is thought to be caused by anomalies in the dopaminergic and cholinergic pathways modulating the striatum.[1,2] Recent studies have also supported the hypothesis that environmental triggers, genetic markers, and susceptibility genes could contribute to the pathophysiology of the disease.[2]

The COVID-19 pandemic developed literature in neurological findings following coronavirus infections. The most reported movement disorder following a COVID infection is myoclonus.[4–6] Other commonly reported hyperkinetic movement disorders are ataxia and opsoclonus.[6] Few studies have reported the occurrence of other movement disorders such as blepharospasm and focal dystonia, but they are thought to be fairly rare.[6] Three cases of blepharospasm have been reported with presentation occurring from 2 weeks to 4 months after the onset of symptomatic COVID infection.[7] One case of dystonia of the upper extremities was described following a symptomatic SARS-CoV-2 infection.[4,8] As for patients already having a diagnosis of dystonia and hemifacial spasm, 65 percent of them noted worsening of symptoms, mostly due to delays in botulinum toxin injections rather than the COVID infection itself.[5] Although the mechanism by which COVID may cause these neurological findings is not clear, most researchers speculate an underlying immunological cause,[4–6,8] such as molecular mimicry, especially since these cases seemed to respond well to immune therapies.[6] In asymptomatic or mildly symptomatic patients, structural damage and toxic or metabolic anomalies are less likely to explain the occurrence of movement disorders, making an immune-mediated cause even more persuasive.[6] Other groups hypothesized a diencephalic dysfunction due to trans-neural penetration of COVID,[5,7] structural damage such as anoxic brain injury or stroke,[4,5] toxic or metabolic anomalies,[6] and adverse drug reactions.[5]

We believe this represents the first reported case of Meige syndrome following COVID infection. Due to the underreported nature of post-COVID movement disorders, it is plausible that the occurrence of COVID acted as an environmental trigger,

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causing Meige syndrome in our patient through epigenetic or immune-mediated interactions. Since hyperkinetic movement disorders following COVID infection have been reported to respond well to immune therapies, corticosteroids and intravenous immunoglobulins might be considered as treatment options. Alternatively, since our patient is in the typical age range of symptom onset and most cases of Meige syndrome are idiopathic, she may have coincidentally developed idiopathic Meige syndrome during the period following her asymptomatic COVID infection. The causal relationship between Meige syndrome and COVID infection is uncertain, but this report adds to the literature of potentially rare manifestations following COVID.

Supplementary material. The supplementary material for this article can be found at https://doi.org/10.1017/cjn.2023.298.

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Ethical statement. This article did not require an IRB approval, as it was a case report. Information was anonymized, and we obtained an informed consent form. We have a signed patient consent form (dated 2022-09-22). We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this work is consistent with those guidelines.

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