Subacute onset dystonia in a woman affected by Parkinson’s disease following SARS-CoV-2 infection

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Abstract

Among the phenotypic spectrum of Parkinson’s disease (PD), dystonia is common, especially in juvenile forms. Dystonic spasms are also observed in patients under dopaminergic treatment or in the case of fever, trauma, surgery, and infections; in a minority of cases dystonia may progress to a severe condition such as status dystonicus [1]. Here we describe the case of a woman affected by Parkinson’s disease who presented severe generalized dystonia, developing in a short amount of time after SARS-CoV-2 infection.

1. Introduction

Among the phenotypic spectrum of Parkinson’s disease (PD), dystonia is common, especially in juvenile forms. Dystonic spasms are also observed in patients under dopaminergic treatment or in the case of fever, trauma, surgery, and infections; in a minority of cases dystonia may progress to a severe condition such as status dystonicus [1]. Here we describe the case of a woman affected by Parkinson’s disease who presented severe generalized dystonia, developing in a short amount of time after SARS-CoV-2 infection.

2. Case report

In April 2020, in the middle of the COVID-19 pandemic in Italy, we evaluated a 58-year-old woman with idiopathic PD, diagnosed when she was 50 years old. The patient, at the onset of the disease, began experiencing a mild tremor in his right hand and left shoulder rigidity which became progressively worse. In about two years the patient was diagnosed with PD alter the tremor had advanced in severity and his left shoulder was becoming increasingly rigid. The patient then began taking dopamine agonists suffered from an important constellation of side effects and then levodopa with good control of tremor and rigidity. At the time of evaluation, when “on” she started to experience “mild” dyskinesias initiated by levodopa dosing and worsening akinetic symptoms while “off”.

The dyskinetic and akinetic symptoms were not so severe and she could work and live an almost normal life.

In the following years, she was treated with different combinations of drugs and clozapine was introduced to control levodopa-induced hallucinations. Recently, antiparkinsonian treatment consisted of levodopa/benserazide and clozapine administration. On April 12, we were contacted by the patient’s husband as her temperature had risen slightly (37.2 °C), and she showed slurred speech and confusion. In the following hours, the patient presented severe spasms of arms and legs. Considering the high risk of contagion, and according to national guidelines, we arranged a session of telemedicine consulting. The patient was sleepy but quickly awakened. Her speech was slurred but intelligible without evidence of dysarthria or aphasia. She was not in respiratory distress (no dyspnea or cough). Blood pressure, heart, and respiratory frequency were within the range of normality. Percutaneous oxygen saturation was 97% in room air. She showed sustained dystonic spasms of the four limbs, reported as persistent in the “off” and the “on” med condition. No other focal neurological deficits were present when compared to the last neurologic examination of two months before. She was advised to increase the daily dose of levodopa (from 400 to 600) and clozapine (from 25 to 50 mg). Two positive nasopharyngeal swab test confirmed the clinical suspect of COVID-19.

The infection had a mild course with fever never exceeding 37.5 °C and temperature dropping after a few days. She never presented respiratory distress or other complications. No therapy for viral infection was prescribed; neurological conditions rapidly improved. At the end of the quarantine, she was evaluated at the Movement Disorder Clinic. Thoracic findings, temperature and oxygen saturation were normal. Neurological examination revealed global bradykinesia, resting tremor of the right hand, and mild upper limbs rigidity. The UPDRS III score in on status was 32/108, and no dystonic spasms of the limbs were present. Within three weeks, the therapy regimen returned to the previous one.

3. Discussion

Neurological manifestations in patients infected with SARS-CoV-2 have been reported, and severe respiratory distress seems to
predispose to neurological disorders. Moreover, patients with advanced PD and longer disease duration are more sensitive to COVID-19 disease [2]. To our knowledge, no case of worsening of dystonic symptoms in Sars-Cov2 infection of PD has been described in the literature. Furthermore, in this case, the severity of the infectious disease was mild, but neurological manifestation was important.

Various mechanisms may explain the CNS involvement by SARS-CoV-2 infection. Encephalopathy may be a consequence of CNS metabolic distress due to fever or hypoxia, in other cases, autoimmune vasculitis or a cytokine storm in the brain or viral direct damage [3] have been pointed out.

In our patient, the short duration of symptoms, their limited clinical relevance and duration may suggest a different origin of transient dystonia. Indeed, an interference of the infection with the levodopa metabolism that spoils its bioavailability within the CNS may be hypothesized. The cell receptor for SARS coronaviruses angiotensin-converting enzyme 2 (ACE2) [4] and the dopamine decarboxylase have been reported to be co-expressed and co-regulated in non-neuronal cell types. As such, the involvement of the synthetic dopamine pathway in the pathophysiology of COVID-19 can be hypothesized [5]. In line with this hypothesis is the observation of improved dystonia with increasing levodopa dosage. Clozapine has also been reported to improve hyperkinetic movement disorders, including tardive dystonia. We prescribed a higher dose of this drug to avoid re-emergence of psychosis, but clozapine may have also played a role in the rapid resolution of subacute limbs dystonia. This case report provides further evidence that the neurological manifestations of COVID-19 not being fully framed. Hence, case reports may be relevant for a better and more comprehensive understanding of the interaction between the virus and CNS.

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Conflict of interests

The authors have no conflict of interest to disclose

Informed consent

The patients signed an informed written consent.

7. Ethical Issues

The case report concerns routine management of a neurological condition during the pandemic. Ethical committee approval was not required.

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