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A case report of acute cervical dystonia after electroconvulsive therapy.

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Reviewer Comments:

Dear authors,
Reviewer #2 has a final request. if you will address his question and revise one last time then I can accept without sending it back out for review

Vaughn McCall

Reviewer #2: The authors have effectively addressed most, though not all, of my concerns. The language used to hypothesize correlation between the onset of the dystonia and the ECT treatment has been softened. However, there remains minimal discussion of the potential role of the anesthesia and anesthesiology team. Cervical dystonia has sometimes been linked to head, neck or shoulder injuries, so the reader wonders whether the anesthetist manually moved the head/neck during the procedure in a way that could have contributed to the dystonia. The authors should describe whether there was any potential correlation between the mechanics of airway management during the ECT treatment and the emergence of the dystonia.

Indeed, manipulation of the neck can be linked tot he onset of acute cervical dystonia. This possible correlation asks for a verification of the course of the second ECT procedure. There was no manipulation of the neck during the second ECT procedure. No head tilt or chin lift was neccesary. Neither was there hyperextension of the neck. Physical therapist examination did not reveal any traumatic injuries.

This was adapted in the manuscript, the absence of manipulation was stressed.

" *During the procedure there was no hyperextension or extreme manupilation of the neck necessary to obtain clear airway. After consultations with a physical therapist*" (L32)

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Conflicts of interest:

None of the authors declare conflicts of interest.

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Dear Sir,

Electroconvulsive therapy (ECT) is a widely accepted and effective treatment for major depression and other psychiatric disorders. Furthermore, treatment gains have been reported in neurological disorders such as Parkinson's disease (PD), neuroleptic malignant syndrome, epileptic seizures, and, proposedly, tardive dyskinesia and dystonia.[1,2] In contrast to the reported beneficial effects of ECT on dystonia, we describe the occurrence of acute post-procedure cervical dystonia.

G., a 73-year-old man was hospitalized because of a first depressive episode with psychotic symptoms. Psychiatric symptoms included severe depressed mood, hopelessness, psychomotor retardation, and psychotic and melancholic features (delusions of doom/demise and financial despair). G. scored 33/44 on the Psychotic Depression Assessment Scale (PDAS), which score is indicative of severe psychotic depression[3]. Although G. had no psychiatric history, his mother as well as two maternal uncles had died due to suicide. In 2019 he was diagnosed with PD. G.'s current medication regimen consisted of acetylsalicylic acid 80mg, atorvastatin 10mg, carbidopa/levodopa/entacapone (37,5-150–200mg), nebivolol 2.5mg, melatonin 3mg, pantoprazole 20mg, valeriana officinalis 500mg, and mirtazapine 15mg, all daily.

Due to the severity of the depression, ECT was started after informed consent was obtained. Using the MECTA SPECTRUM 5000Q device (MECTA corporation, OR) we opted for right unilateral d'Elia (temporoparietal) electrode position, with the anesthesia consisting of etomidate 14mg, suxamethonium chloride 15mg and nicardipine 2mg. Stimulus dose was 224mC (with a pulse width of 0.5 milliseconds, a frequency of 35Hz and stimulus duration of 8 sec-

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onds) using the half-age stimulation strategy. Following the second ECT procedure, the patient developed acute cervical dystonia. Dropped head syndrome was observed in the recovery room immediately after procedure. Rigidity of the neck was noted and described as painful. Passive

retroflexion of the neck only slightly possible. Cortical myoclonic jerks at the upper limbs were observed. Right unilateral extrapyramidal image was seen. This acute manifestation of cervical dystonia increased patients depressed mood and feelings of hopelessness. During the procedure there was no hyperextension or extreme manipulation of the neck necessary to obtain clear airway. After consultations with a physical therapist and neurologist, CT imaging was performed, revealing cervical discarthrosis, osteophyte/disc bulging, and degenerative stricture (C4-C7), for which bilateral infiltration with xylocaïne 1% and methylprednisolone acetate 40mg was given and physical rehabilitation was started, without amelioration. The treatment was augmented with levodopa, again without any effects on the dystonic symptoms. Biochemical examination and EMG did not reveal any abnormalities suggestive of myopathic disease. There was no laboratory evidence for hyperparathyroidism. Trial treatment with pyridostigmine-bromide did not improve the dystonia, while the screen for myasthenia antibodies was negative. In conclusion, no clear etiology was found for this acute manifestation of cervical dystonia.

After consulting the patient and his family, it was decided to continue ECT given the severity of the depression, the hopelessness, and suicidality. The ECT course eventually comprised a total of 11 treatments with doses being increased up to 352mC. After the sixth session, mood and psychomotor symptoms first started to show improvement and full clinical remission was achieved after the 11th session (PDAS 7/44), following which weekly continuation ECT was started. Although half-age stimulation strategy is not the ideal treatment in right unilateral d'Elia electrode position, adequate seizures were evoked and remission was achieved. The dystonia had improved over the course of the ECT sessions, but symptoms

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remained. Less rigidity of the neck was noted, pain subsided. Also more passive retroflexion was possible.

There was no clear cause found for this acute dystonia. Its presentation immediately after the second session raises the question whether there could be a correlation between the ECT treatment and the acutely developed dystonia. Verification remains difficult, however.

To our knowledge, Sienaert and Peuskens[4] are the only authors to date to have reviewed published case reports concerning the impact of ECT on dystonia. In their case report of 2005, they found six cases to discuss beneficial effects of ECT on dystonia, one to report no post-ECT improvement and one an adverse effect. However, in the latter case the cessation of antipsychotics prior to ECT could have caused withdrawal dyskinesia, thus partially explaining the worsening of motor symptoms[5]. In our case no medication alterations were made, ruling out withdrawal dyskinesia. Also etomidate seems an unlikely cause. Although the anesthetic agent has been described to induce transient muscle movements, we found no evidence in the literature describing acute cervical dystonia as an adverse effect.

The comorbid presence of PD makes our case complex. Individuals with PD are more vulnerable to develop (acute) dystonia due to a lack of dopamine, but based on the current literature they are not considered to be more susceptible to developing dystonia as a result of ECT. On the contrary, ECT has been shown to improve motor symptoms in PD[2]. Some reports even suggest that in PD higher symptom severity and older age are associated with a better motor response to ECT. Still, while PD is not taken as a contraindication for treatment with ECT[2], patients with depression and PD appear to be more sensitive to side effects (mainly delirium), making close monitoring during treatment even more essential.

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We are the second to report the occurrence of acute dystonia after ECT, contrasting earlier case reports describing post-treatment improvements. A rare event, it does pose a serious dilemma: do we halt or continue ECT? Considering the severity of our patient's depressive and comorbid symptoms, it was decided to continue the ECT course, which resulted in remission of his psychotic depression. Also, dystonic features mildly improved. However, it remains difficult to correlate both the development and the subsidence of the dystonia with the ECT treatment. Knowledge concerning the underlying mechanisms of both dystonia and ECT treatment remains limited. Although ours and other case reports add to the literature, further research is needed to better guide clinicians.

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