Title: Atypical Presentation of Neuroleptic Malignant Syndrome in a Patient with Autism Spectrum Disorder Requiring ECT

Background/Significance: There is a known relationship between catatonia and neuroleptic malignant syndrome (NMS), with many authors believing that it represents a single spectrum of illness (Fricchoine, 1985). Patients with autism spectrum disorder (ASD) may have catatonia-like features at baseline (Klek et al., 2023). Patients with ASD who are diagnosed with NMS may have physical exam findings that reflect both syndromes. In this report, we present a patient with ASD who presents with NMS that ultimately required ECT.

Case: Mr. L is a 29-year-old male with a history of ASD, ADHD, and depression who presented with altered mental status and tachycardia. He had increasing agitation prior to admission, resulting in escalating doses of risperidone and haloperidol over three weeks. His baseline physical exam prior to hospitalization was unobtainable. He then developed significant fever, tachycardia, elevated CK, and lead-pipe rigidity, resulting in a diagnosis of NMS. He was initially given benzodiazepines, bromocriptine, and dantrolene as well as cyproheptadine. His condition then worsened, requiring intubation. ECT was quickly initiated for refractory NMS even while workup was still ongoing. MRI, lumbar puncture, and EEG were benign. Tachycardia and fever improved, and rigidity gradually subsided. Initial presentation of ankle clonus and hyper-reflexia did not resolve throughout the nine treatments of ECT, and were thought to be part of his baseline exam. The patient was discharged one week after stopping ECT.

Discussion: ECT has long been indicated for cases of refractory NMS after failure of pharmacotherapy. Although significant differences in mortality have not been shown across all cases of NMS, treatment with ECT had 0% mortality, while bromocriptine and dantrolene had 8.5%, and supportive care alone showed 10.2% mortality (Kuhlwilm 2020). This highlights the necessity of urgent ECT for patients with refractory NMS, even at the expense of some diagnostic workup. Our patient initially showed improvement of neurologic function by the 3rd ECT, with significant progression and return to verbal status by the 6th ECT. The efficacy of ECT for this patient would have been even more definitive if the team had the opportunity to obtain a baseline exam.

Conclusion: ECT was shown to be an effective treatment for this patient with ASD who presented with NMS and atypical physical exam. This case shows that medical teams should be mindful of exam findings that could be red herrings, and their workups should not delay the transition to ECT. Having a baseline exam would help limit the need for extraneous workup.

References:

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