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A rare case report of Gjessing's syndrome - Periodic catatonia

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Abstract
Background*: Here we describe, Master x, a 17 year old male who had presented to the Psychiatry out-patient department (O.P.D) with abrupt onset of withdrawn behaviour, being less communicative, stupor and fearfulness, with a previous history of two such episodes at irregular intervals in the past illustrating that periodic catatonia can still occur as an independent disorder, warranting a need to relook into the current classification.*

Keywords: *periodic catatonia, gjessing syndrome, catatonia*

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I. Introduction

Gjessing syndrome was identified by Rolv Gjessing and Leiv Gjessing in 1925 with the features of catatonic symptoms recurring in an individual. From the series of the cases reported the patients were mostly young males who have onset of symptoms in the age of 14 – 15 years with a periodic recurrence of symptoms. [(1)] Periodic catatonia is frequently underdiagnosed because of its episodic presentation with near complete resolution in between the episodes. The prevalence of catatonia is unknown. Catatonia is likely underdiagnosed by psychiatrists and other physicians. It is common among psychiatric inpatients (7%–31% in six prevalence studies conducted after 1976 [total N=1,081]), and it occurs most often in patients with mood disorders (28%–31% of catatonic patients had mixed mania or mania in three studies conducted since 1977 [total N=280]). [(2)] The lifetime prevalence of periodic catatonia which was estimated at 0.001 in the general population. [(3)] In recent advances into identifying the aetiology studies done by Northoff proposes that the initial neurochemical dysregulation in catatonia occurs in the cortex, with involvement of the GABA-A system and subsequent dopamine dysregulation in the thalamocortical loops and subcortex [(4)].

II. Case Report

A 17-year-old male was brought to the O.P.D by his father and mother with complaints of withdrawn behavior, fearfulness, increased frequency of micturition and decreased sleep. He had his first episode in 2018 during his 10th std exams when he complained of fearfulness, was withdrawn to self and subsequently he became mute, was unable to respond adequately to the verbal instructions and had to be coaxed to eat food and had sleep disturbance then.

His parents initially sought magico -religious treatment and later consulted a private psychiatrist. The episode lasted for 15 to 20 days duration and the parents reported that he returned to normal functioning and was maintaining relatively well. He completed his exams and joined 11th std and continued attending classes.

He was functioning relatively well until August 2020 when he again started becoming fearful and withdrawn. On further probing, his parents reported that he told them that he could see blood dripping from his arms and hairs all over on the floor. In addition, he could see some weird images that were not seen by others. This episode lasted for approximately same duration like the previous one. Due to covid they could not access treatment, but the symptoms resolved spontaneously. He gradually started interacting with others and resumed near normal functioning within 20 days.

He had his third episode on the 22nd of July 2021 which was acute in onset. He started talking less with decreased food intake and had sleep disturbance. The following day he developed fever and had one episode of giddiness which was not associated with any involuntary movements or other features suggestive of seizure semiology or organicity. He was appearing fearful and was clinging to his mother.

He was neither sleeping nor talking adequately. But, during these episodes the parents reported that he would listen to whatever they say without any resistance like. They also told that he would drink a lot of water

and frequently used the restroom. On the day before coming to the O.P.D he had an episode of urinating in the bed during night and there was no history of similar complaints in the recent past.

PAST HISTORY: No history of comorbidities, head injury, or taking regular medication

FAMILY HISTORY: He is the eldest son and has a younger sister has history of febrile seizures.

PERSONAL HISTORY: Normal delivery, breastfed, fully vaccinated no h/o birth complications or developmental milestone delay reported. No history of behavioral issues reported with below average scholastic performance. No h/o substance use reported.

TEMPERAMENT: Parents describe him as an introverted child with reduced social interaction. He is easily adaptable and distractable. He is slow to warm and avoids conversations with strangers. He spends his leisure time playing games in his phone.

ON EXAMINATION: He was thin built moderately nourished
Blood pressure was 120/80. Pulse rate: 90bpm Weight: 38 kgs

MSE:

He was well dressed and kempt. His facial expression was dull with a vacant stare. Eye contact was not made or maintained. Rapport could not be established. He remained mute and did not respond to any questions asked. He was aware of the surroundings and responded to environmental cues. His psychomotor activity was decreased during the interview. He frequently wanted to use the restroom. On further examination he had ambitendency, automatic obedience, echopraxia and posturing. On examination, he voided urine without any resistance though he had clear consciousness and was alert suggesting urinary retention, a rare phenomenon seen in catatonic individuals. Bush Francis rating score was 24.

Investigations:
2018: EEG: Normal study

2021:

COMPLETE BLOOD COUNT
Hemoglobin: 9.89 grams/dL
R.B.C: 3.7 trillion cells/L
Total count:7200 cells/mcL

RENAL FUNCTION TEST:
Urea: 21.15 mg/dl
Creatinine: 0.5 mg/dl
Uric acid: 10 mg/dl

SERUM ELECTROLYTES:
Sodium:141mEq/L
Chloride:97.55 mEq/L
Potassium: 3.8 mEq/L

LIVER FUNCTION TEST:
Bilirubin:0.9 mg/dl
Sgot:20.84 IU/L
Sgpt:24.44 IU/L
Alkaline phosphatase: 87.34
Total protein:7.04 gm/dl
Albumin:4 gm/dl
Globulin:3.4 gm/dl
Albumin globulin ratio: 0.85
GGT: 32

THYROID FUNCTION TEST:
T3: 3.47 ng/ml
T4:1.36 µg/dL
TSH: 2.15 mIU/L

III. Treatment

The mainstay of treatment of catatonia has been benzodiazepines. Ever since Huang et al [(5)] reported the use of a lorazepam – diazepam protocol to relieve catatonic symptoms in schizophrenic patients, it has been proven to be an efficient treatment. Lorazepam challenge at a dose of 1mg diluted in 4ml of normal saline was administered and within half hour he started to write. The patient was started on started on treatment with clonazepam 0.5 mg morning and afternoon and one mg at night for 5 days initially and then switched to lorazepam 2mg. He was also started on olanzapine 5mg od which was increased to B.D on follow up. On the next follow up mother reported that the fearfulness reduced. Dosage was increased further and on subsequent follow ups his fearfulness decreased significantly and he was interacting better. He was smiling ambitendency

and automatic obedience could not be elicited. He was interacting with the family members and the patient was having spontaneous speech and self-care.

IV. Discussion

Our patient experienced periodic catatonic episodes that remitted spontaneously initially with minimal residual symptoms. Initially Gjessing described this disorder of periodic catatonia, and he systematically studied the metabolic disturbances in these patients. He suggested that the behavioural fluctuations of periodic catatonia were related to a cyclic nitrogen imbalance and could be treated with thyroid hormone extract, which seemed to control the symptoms but not cure the disorder [(6)] .Adding to this, Kahlbaum conceptualized catatonia to have a cyclical and alternating course which was evident in our case. The case presented here had subtle qualitative changes with each subsequent episodes as described by Leonhard in his classificatory system which reported that patients with periodic catatonia tend to displayy qualitative changes in addition to the quantitative changes during acute episodes with accumulating defects after repeated episodes [(7)] .

ICD-10 still categorizes catatonia under schizophrenia, whereas in DSM-5, it is no longer a subtype of schizophrenia as it includes an unspecified category of catatonia. This is substantiated by literature which reported that approximately 4 - 46% of patients diagnosed with catatonia had no known identifiable underlying condition [(8)] .

Our case reported here had one fever spell with autonomic instability and urinary retention. Presence of fever with autonomic instability is a well-established risk factor for malignant catatonia which has a death rate of 9%. [(9).]

The urinary retention described in our case could be considered as an extreme form of negativism. Both the above-mentioned symptoms required urgent intervention. Hence accurate diagnosis and treatment of catatonia is crucial in clinical practice. It may not only avoid somatic complications but also the development of resistance to treatment as well.

Though catatonia is more common than thought, research studies investigating the prevalence, pathophysiology, and treatment of this syndrome are sparse. Periodic catatonia is generally considered as a rare heritable subtype of catatonic schizophrenia with a chronic degenerating course [(10)] . Our patient experienced periodic catatonic episodes that remitted spontaneously initially with minimal residual symptoms.

V. Conclusion

Psychiatrists are in a position to diagnose and treat catatonia on both medical and psychiatric units, given the combination of medical and psychiatric problems that catatonic patients often present. Though catatonia is more common than thought, research studies investigating the prevalence, pathophysiology, and treatment of this syndrome are sparse. Till date Benzodiazepines and ECT continue to be mainstays of treatment, and evidence is mounting for the use of NMDA antagonists in catatonia refractory to lorazepam. The safety and usefulness of atypical antipsychotic medications in catatonia due to a general medical condition remain unclear, though there appears to be some evidence for their effectiveness in patients with catatonic schizophrenia. An increased awareness of the diagnosis and treatment of catatonia among clinicians has the potential to significantly improve morbidity and mortality of catatonic patients. Further research into its pathophysiology and clinical treatment will offer us an excellent chance at expanding our understanding of psychiatric illness and its causes.

References

[1]. Gjessing R, Gjessing L. Some main trends in the clinical aspects of periodic catatonia. Acta Psychiatr Scand. 1961;37(1):1-13. doi: 10.1111/j.1600-0447.1961.tb06155.x. PMID: 13899055.

[2]. Catatonia: Clinical Aspects and Neurobiological Correlates Jessica Daniels M.D.Published Online:1 Oct 2009

[3]. Solmi M, Pigato GG, Roiter B, et al. Prevalence of Catatonia and Its Moderators in Clinical Samples: Results from a Meta-analysis and Meta-regression Analysis. *Schizophr Bull*. 2018;44(5):1133-1150. doi:10.1093/schbul/sbx157

[4]. Northoff G: What catatonia can tell us about "top-down modulation": a neuropsychiatric hypothesis. Behavioral and Brain Sciences 2002; 25:555–604

[5]. Chin-Chuen Lin, Yi-Yung Hung, Meng-Chang Tsai, Tiao-Lai Huang, The Lorazepam and Diazepam Protocol for Catatonia Due to General Medical Condition and Substance in Liaison Psychiatry, PLOS ONE, 10.1371/journal.pone.0170452, **12**, 1, (e0170452), (2017).

[6]. Fink M, Taylor MA: The many faces of catatonia, in Catatonia: A Clinician's Guide to Diagnosis and Treatment. New York, Cambridge University Press, 2003, pp 59–60

[7]. Caroff SN, Hurford I, Bleier HR, Gorton GE, Campbell EC. Recurrent Idiopathic Catatonia: Implications beyond the Diagnostic and Statistical Manual of Mental Disorders 5th Edition. *Clin Psychopharmacol Neurosci*. 2015;13(2):218-221. doi:10.9758/cpn.2015.13.2.218

[8]. Fink M, Shorter E, Taylor MA. Catatonia is not schizophrenia: Kraepelin's error and the need to recognize catatonia as an independent syndrome in medical nomenclature. *Schizophr Bull*. 2010;36(2):314-320. doi:10.1093/schbul/sbp059

[9]. Rasmussen SA, Mazurek MF, Rosebush PI. Catatonia: Our current understanding of its diagnosis, treatment and pathophysiology. *World J Psychiatry*. 2016;6(4):391-398. Published 2016 Dec 22. doi:10.5498/wjp.v6.i4.391

[10]. Edinoff AN, Kaufman SE, Hollier JW, et al. Catatonia: Clinical Overview of the Diagnosis, Treatment, and Clinical Challenges. *Neurol Int*. 2021;13(4):570-586. Published 2021 Nov 8. doi:10.3390/neurolint13040057