CASE REPORT

Brief episodes of non-specific psychosis later diagnosed as periodic catatonia

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SUMMARY

A 73-year-old woman was known to have discrete episodes of psychosis not otherwise specified that would require a brief admission to hospital and total remission following a short course of benzodiazepine or antipsychotic treatment. She had no underlying schizophrenic or affective disorder and was completely unimpaired in between episodes, which could last several years. She presented to us with psychotic symptoms but also noted to have many catatonic features, which were also present on previous presentations. Following failure with antipsychotic trials on this index presentation, she completely remitted with a short course of electroconvulsive therapy.

We discuss the importance of identifying and treating catatonia and the lesser-known variant of periodic catatonia. Current presentations should always take into account the lifetime context of psychiatric illness. Rarely do patients with primary psychotic disorders not have any impairment or treatment in between episodes.

BACKGROUND

Catatonia has been associated with many psychiatric and medical conditions and generally is thought as a condition that is secondary to a primary major psychiatric disorder.[1] The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) uses catatonia as a possible specifier for a number of conditions, and established a diagnosis of catatonia not otherwise specified (NOS), used when the underlying cause is not yet identified. There has been a long history of advocating for the establishment of catatonia as a disorder of its own.[2] Studies suggest that diagnoses of catatonia are commonly found with no known underlying condition, referred to as idiopathic catatonia.[3 4] A forgotten subtype of catatonia that previously generated clinical and research support is periodic catatonia,[5] characterised by rapid onset, brief and recurrent episodes of catatonia with a longitudinal course. Prognosis is typically better than systematic catatonia, which is insidious and progressive. Gjessing[6 7] described periodic catatonia in great detail through a comprehensive series of case reports, where episodes could be separated by years. Here we report a case of periodic catatonia throughout a patient's lifespan without any underlying psychiatric condition, including schizophrenia or major affective disorder. This case illustrates the diagnostic confusion around catatonia, which if properly identified can respond extremely well to electroconvulsive therapy (ECT) as in our patient.

CASE PRESENTATION

A 73-year-old woman presents with an acute history of increasing mutism, immobility and withdrawal. She would not eat, drink or participate in self-care. Earlier in the course of this presentation, she would have periods of overt confusion, disorientation and varying intensities of anxiety, paranoia, delusions and hallucinations. According to her family, she has had patterns of becoming acutely ill for the last 45 years. This would involve a consistent pattern of stupor, withdrawal and confusion, mixed with periodic bouts of psychotic symptoms or mood disturbances. Typically, these cases would resolve promptly with hospitalisation and treatment with lorazepam and/or an antipsychotic. Remarkably, she would be completely well in between these episodes and would not need any psychiatric follow-up. Her only current psychotropic medication was olanzapine, which she was started on in her last admission, 15 years ago. She has been on this medication and without any psychiatric contact. Records from an admission 17 years ago stated she achieved remission to a similar episode only with scheduled lorazepam treatment. Those notes also mention that her older admissions had similar presentations that would resolve with a short course of chlorpromazine and then discontinued. At those times, she was thought to be having either a non-specific psychotic episode or depression with psychotic symptoms. There has been no consistent pattern of triggers that precede these episodes. Medical history only noted remote surgeries in her leg for a motor vehicle accident. There was no family history of any major psychiatric disorders.

Initially, she was treated here as having a non-specific psychotic episode with catatonic features. Eventually, catatonia was recognised as the predominant syndrome, presenting with many tell-tale signs of catatonia, including echolalia, mutism, stupor, staring, rigidity, negativism, withdrawal and gegenhalten.

DIFFERENTIAL DIAGNOSIS

This patient presents as a unique case for a number of reasons. First is the recurrent, episodic nature of the illness and being essentially asymptomatic in between. She was also not on any medications between her episodes, with the exception of the period in between the index admission and the previous one (age 56 to the present, 71). During those years, she was on olanzapine, which may have been unnecessary, as there was no identifiable indication. Episodic psychosis typically points towards an

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exacerbation of schizophrenia or bipolar illness, but these disorders occur on the background of ongoing debilitation, either through persisting executive dysfunction, residual depression and anxiety, or psychosocial impairment. Even if patients with bipolar disorder or schizophrenia are asymptomatic, they should require ongoing maintenance treatment. Moreover, both of these illnesses typically present with psychiatric illness in the family history, and withdrawal of medications, as in her earlier history, should exacerbate symptoms. Her diagnoses given in previous admissions have been dissociative disorder or psychosis NOS. Dissociation typically presents with an acute onset of disorientation, depersonalisation and amnesia. Dissociative amnesia is a possible diagnosis, but our patient lacks the characteristic association with psychological trauma. Moreover, dissociative disorders are not typically recurrent in a stereotyped fashion, and response to biological therapies is not robust. Although primary psychotic disorders like schizophrenia, bipolar disorder and psychotic depression present with several indices of cognitive impairment, frank loss of orientation and memory is not seen. Most importantly, our patient exhibited the psychomotor signs associated with catatonia as the central presentation, which included stupor, rigidity, mutism and staring. On the basis of the presentation and differential diagnosis, our impression was that this patient suffered from idiopathic catatonia, which is a condition without any other underlying psychiatric illness.

TREATMENT

She was initially switched to aripiprazole and haloperidol, titrating up to 14 and 5 mg, respectively, over 1 month with no results. She was then tapered off of these medications, and quetiapine was initiated. She remained mostly unresponsive. She was started on ECT but did not attain adequate seizures in the first two sessions. She was switched off of her quetiapine and back onto olanzapine and titrated up. After two more sessions of ECT, she started to improve dramatically. She became increasingly alert, responsive and eating her meals. She could not remember any of the details or events leading up to her presenting illness. She denied any depression, anxiety, hallucinations or delusions. After another two sessions of ECT, these behaviours began to subside. By eight treatments over 3 weeks, she had returned to her baseline, premorbid state.

OUTCOME AND FOLLOW-UP

She remains in remission 12 months following discharge.

DISCUSSION

Catatonia is a unique syndrome that responds to specific treatments; first with benzodiazepines and second with ECT. Including catatonia in the differential can have a significant influence in management. ECT has good efficacy in catatonic patients.[7] Rapid response can be expected, and the American Psychiatric Association states that it is the most effective treatment for catatonia.[8] Better response is associated with younger age, longer seizure duration, more severe vegetative impairment and earlier initiation of ECT.[9] Thus, it is important that ECT be considered when catatonic signs can be identified in an episode of psychosis or otherwise.

Some reports in the literature advocate for antipsychotics not to be used, due to evidence that they can exacerbate catatonia and have increased risk of neuroleptic malignant syndrome.[10] Contrary evidence exists, however. A case report described a patient with periodic catatonia that recovered from her periodic episodes with olanzapine and clonazepam.[11] Caroff and colleagues[12] report a case that did not respond to adding ECT to lorazepam, but eventually remitted on haloperidol. Another case describes a catatonic man with a history of major depression, schizoaffective disorder and post traumatic stress disorder not responding to lorazepam but did improve with perphenazine.[13] Thus, perhaps they should only be used in the context of a specific underlying psychotic disorder and not otherwise. Indeed, catatonic signs in patients with first episode psychosis responded very well to antipsychotic monotherapy, and this effect was entirely dependent on the effect of medication on positive psychotic symptoms.[14] With our patient, it is unlikely that the olanzapine contributed to her clinical improvement, as it had been a stable medication of hers for a long time prior to and during initial phases of treatment before it was re-introduced. As described earlier, her presentation also does not fit with a schizophrenic catatonia. Krishna and colleagues[3] argued that patients presenting with a lesser duration and severity of psychotic symptoms contrasted with a much heavier burden of catatonic signs would be in keeping with an idiopathic catatonia, which is in keeping with our patient's presentation.

A pertinent issue in the catatonia literature is underrecognition of the syndrome.[10] Studies have shown that prevalence is about 10% of inpatients[15] and is commonly under diagnosed because clinicians do not routinely screen for these symptoms and signs.[16] With this patient, previous discharge summaries make no note of catatonic signs, but she and her family all report that each of her episodes was characterised by withdrawal, mutism, stupor and rigidity. This suggests that screening for catatonic signs may not be routinely addressed in clinical practice, thus delaying adequate diagnosis and treatment. Moreover, recognition of another underlying psychiatric disorder such as schizophrenia or major depression is not required. Although idiopathic catatonia is not recognised in the DSM classification, catatonia as a distinct syndrome has been described extensively[2] and indeed was the original conceptualisation of catatonia.[1] Karl Kahlbaum was the first to describe catatonia in his monograph as a syndrome of cycling through symptoms of mania, melancholia, confusion and dementia, but the motor signs were the underlying characteristic features pointing to a diagnosis that he stated was a discrete entity.[17] More modern studies suggest that diagnoses of catatonia are commonly found with no known underlying condition, ranging between 4% and 46% of cases.[16]

Learning points

- Catatonia is a psychomotor syndrome that is frequently overlooked and under-reported.
- Catatonia can be diagnosed and treated without another underlying psychiatric disorder. More commonly described diagnoses of schizophrenia, bipolar disorder or major depression may not be a good fit after closer examination of the patient's longitudinal history. In these cases, the patient may have an idiopathic catatonia that is still amenable to treatment.
- Periodic catatonia occurs when patients have brief, recurrent episodes of catatonia throughout the lifespan.
- Benzodiazepines and ECT are specific treatments for catatonia. The latter is reserved as second-line treatment, but may be the most effective treatment.
- Antipsychotics may be ineffective or worsen the condition of catatonic patients. In these cases, clinicians should question if there is truly a primary psychotic disorder present.

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Contributors VMT and HP were involved in the clinical care of the patient, writing and editing of the manuscript and obtaining informed consent.

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