**Refractory catatonia in old age: a case report**

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**Abstract**

**Background:** Catatonia is a clinical syndrome characterised by psychomotor disruption, which often goes undiagnosed. Most reports have focused on interventions and outcomes for catatonia in younger people and those with schizophrenia. The clinical characteristics and course of catatonia in old age is poorly understood. We present a report of an older person whose catatonia was refractory to extensive treatment, and we identify important implications for the management of catatonia in old age.

**Case presentation:** We describe a 73-year-old man with longstanding autistic spectrum disorder who presented with symptoms of depression. Following a period of diagnostic uncertainty and failure to improve with antidepressant medication, a lorazepam challenge yielded an abrupt improvement in presentation. The patient was treated extensively with lorazepam, zolpidem and electroconvulsive therapy during his 16-month hospital admission, but his catatonia ultimately proved refractory to treatment.

**Conclusions:** Catatoniashould be considered promptly as a differential diagnosis in older people presenting with atypical features of functional mental illness. Although partial improvement of catatonic features was achieved using benzodiazepines and electroconvulsive therapy, these were not sustained in our patient. We identified co-morbid autistic spectrum disorder, prolonged duration of catatonia and sensitivity to benzodiazepines as important factors in prognostication in old age.

**Background**

The first descriptions of catatonia can be traced back to 1849 when Bell reported on 40 patients presenting with concurrent mania, delirium, psychosis, sleeplessness and over-activity (1). In 1874 Kahlbaum described catatonia as an independent psychiatric syndrome characterised by a cyclical course of alternating manic, depressive and psychotic phases, with eventual deteriorating course (2). Whilst catatonia has historically been most closely associated with schizophrenia, it has more recently been recognised as a syndrome related to a range of psychiatric and medical disorders (3).

Catatonia is now defined as a clinical syndrome characterised by the co-existence of psychiatric and motor signs (4). Criteria for catatonia are presented in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5), and summarised in **Table 1**. Timely recognition of catatonia can be challenging, possibly due to poor awareness amongst some clinicians, or the longer period of observation often required for some catatonic signs to emerge (5). Untreated catatonia can lead to a range of recognised complications including infection, venous thromboembolism, aspiration pneumonia and even death (6). Catatonia is, however, very much a treatable condition with a response rate thought to range between 59% in patients with schizophrenia to over 90% in patients with other psychiatric diagnoses (4).

**Table 1: Clinical diagnosis of catatonia**

|  |  |
| --- | --- |
| Catalepsy | Passive induction of a posture held against gravity |
| Waxy flexibility | Slight and even resistance to positioning by examiner |
| Stupor | No psychomotor activity; no reactivity to environment |
| Agitation | Agitation, not influenced by external stimuli |
| Mutism | No, or minimal verbal response |
| Negativism | Opposing or not responding to instructions or external stimuli |
| Posturing | Spontaneous and active maintenance of a posture against gravity |
| Mannerisms | Odd caricature of normal actions |
| Stereotypies | Repetitive, abnormally frequent, non-goal directed movements |
| Grimacing | Maintenance of odd facial expressions |
| Echolalia | Repeating the words spoken by the examiner |
| Echopraxia | Mimicking the movements made by the examiner |

DSM-5 criteria for catatonia (7), defined as the presence of three or more of the clinical features listed above.

Benzodiazepines are the mainstay of treatment of catatonia (4, 6), with lorazepam the most used drug at a dose between 2-16 mg/day (8). Indeed, catatonia can be confirmed with a lorazepam challenge test, where the patient is examined before and after a dose of 1-2mg of lorazepam. A positive response is a swift and marked reduction in catatonic signs (6), which can be quantified using the Bush-Francis Catatonia Rating Scale (9). Whilst the precise mechanisms underlying the pathophysiology of catatonia are still poorly understood, it is hypothesised that it may be characterised by gamma-aminobutyric acid (GABA) receptor cortical dysregulation and deficits in the dorsolateral prefrontal cortex (10). It is therefore postulated that benzodiazepines may act by increasing cerebral GABA signalling. Where treatment with lorazepam is not successful, or when adverse effects prevent use of therapeutic doses, electroconvulsive therapy (ECT) can also be highly effective (8).

Early recognition and treatment of catatonia generally leads to rapid resolution of symptoms (6). However, the presentation, management and prognosis of catatonia in old age is poorly understood with only a handful of case studies published in this field to date (11-13). In this report, we describe the case of an elderly catatonic patient who was ultimately refractory to treatment, raising important implications for the management of catatonia in old age.

**Case presentation**

This 73-year-old white man had not previously been known to specialist mental health services and in fact not seen his general practitioner (GP) for over three decades. His family reported that he had periods of depression during his late teenage years, when he left the merchant navy after only a few weeks, and in the third and fourth decade of his life. He neither sought nor received any treatment for these episodes. He never married, had no children and did not work after leaving the navy. He lived alone following the death of his co-habiting parents and relied heavily on his siblings and nieces for assistance with food shopping and house maintenance. Notably, his mother had a history of recurrent depression with inpatient psychiatric admissions requiring ECT.

The patient’s family report him having always been different in personality. He struggled in social settings, found it hard to make friends and had never been in a romantic relationship. He was obsessional about routine and was known as a child to pace and exhibit repetitive actions when anxious. He also had very specific and intense interests, such as obsessively learning the Latin names of plants. He had not previously been diagnosed with a childhood or developmental mental disorder. His only medical history consisted of a tonsillectomy and childhood scarlet fever. He took no regular medication, did not smoke cigarettes, consumed no alcohol and there was no recreational drug use. He was not known to the social care service and had no forensic history.

At the start of the current episode, the patient was referred for an urgent mental health consultation by his GP due to concerns about his mental state over the preceding few weeks. He had retreated into his bedroom and was not washing himself or eating. He was noted to be unkempt, thin and frail. He displayed objective signs of depression and reported feeling lonely. He stated that he had suicidal thoughts but denied intent. He was noted to speak quietly but was repetitive with short responses. He was not fully orientated to time or place, and his attention was notably poor when tested. He was admitted to a psychiatric ward for older people for a period of assessment. His GP found no acute physical cause for his symptoms and on admission no abnormalities were detected on all initial investigations, including a battery of blood tests. There were no positive findings on physical examination and specifically no focal neurology. His observations showed hypertension with a blood pressure of 160/72 mmHg, but were otherwise unremarkable.

A long inpatient admission began, which would eventually last 16 months as summarised in a timeline shown in **Figure 1**. Despite being admitted informally, he was soon deemed to lack mental capacity to consent to admission and was detained under the UK Mental Health Act 1983 (amended 2007). His clinical presentation included tearfulness, poor eye contact, reduced content of speech, frequent pacing, expression of thoughts of self-harm and mimicking stabbing motions on himself. He wrote about a wish to die in his diary on the ward, although the content of this was disjointed. Collateral history from his family was received regarding his premorbid personality and level of functioning. It was concluded that he was experiencing severe depression on a background of autistic spectrum disorder. He was commenced on mirtazapine and then risperidone which were titrated to effect, with sertraline added as a second antidepressant. During his initial assessment the diagnosis of hebephrenic schizophrenia was considered. This was excluded after his family confirmed that prior to the current episode he had functioned relatively well, showering independently, and preparing his own meals for example, and his speech and behaviour were not disorganised.

During the early weeks of his admission the patient progressively became mute. He attempted to eat inanimate objects such as soap bars and continued to pace relentlessly. He also became intermittently incontinent of urine and faeces. He was reviewed by the department for neurological sciences who excluded conditions such as motor neuron disease, and a negative auto-antibody screen helped to exclude a diagnosis of limbic encephalitis. Dementia was suspected and a brain computed tomography (CT) scan was attempted, but the patient did not tolerate this. Due to the importance of excluding an organic brain disorder, a repeat CT brain scan was organised but this time using conscious sedation with oral Lorazepam (2mg pre-scan). This resulted in an unexpected, sudden, and dramatic clinical response within minutes. The patient began speaking fluently and coherently for several hours, until his presentation of mutism, increased motor activity and agitation returned. The use of Lorazepam for conscious sedation, and the subsequent clinical response, was interpreted as a positive Lorazepam challenge and a syndrome of catatonia was confirmed. A trial of 0.5mg Lorazepam four times per day was commenced, however, the dose was reduced and then stopped altogether within days due to severe sedation and frequent falls.

Following the unsuccessful trial of oral Lorazepam, our patient was commenced on bilateral, twice weekly ECT (Somatics Thymatron System IV). After 9 treatments there were some signs of improvement, with brief and short episodes of speaking and increased writing in his diary. With ongoing ECT his mutism became more intermittent and this revealed disordered thought form. A diagnosis of schizophrenia was revisited as a possible contributing factor to his catatonia, with risperidone replaced by aripiprazole. Although still showing some signs of improvement after 24 ECT treatments, the patient continued to constantly pace and remained mute most of the time.

Notably, CT brain imaging and later magnetic resonance imaging (MRI) findings included mild generalised cerebral atrophy, including mild bilateral hippocampal volume loss, but no specific features of neurodegenerative disease, Creutzfeldt-Jakob disease or cerebral amyloid angiopathy.

Following continued failure to significantly improve with ECT, antipsychotic medication was suspended due to its possible effect on the poor response to ECT in catatonia, as has been reported previously (14, 15). Sertraline and mirtazapine were also stopped to exclude serotonin syndrome as a potential cause of his restlessness. Zolpidem, which has previously been suggested as a treatment for catatonia (16), was then trialled for 2 weeks but provided no benefit. Following completion of 35 treatments of ECT, the patient remained mostly mute and displayed continued psychomotor agitation. However, his presentation then changed markedly to become rigid and unresponsive to external stimuli, demonstrating stupor. He continued to be mute and demonstrated repetitive teeth clenching (stereotypy) with negativism. He scored 16 on the Bush Francis Catatonia Rating Scale. This deterioration in presentation prompted a further trial of lorazepam, initially given intravenously under close monitoring in a local general hospital.

As before, the first dose of lorazepam provided marked but temporary improvement in symptoms, as the patient started to talk fluently, and his rigidity disappeared. He was again trialled on regular intravenous then oral lorazepam, cautiously increased from 0.5mg twice daily to 6mg daily. Unfortunately, the patient developed aspiration pneumonia and remained in the general hospital for treatment of this. His prognosis was thought to be very poor and his lorazepam was stopped due to the risk of respiratory depression. He was deemed too unfit for anaesthetic to undergo further ECT. At this point he was transferred back to our psychiatric hospital for end of life care, with his family in agreement.

Somewhat remarkably, shortly after transfer he became slightly more alert and was able to be fed by staff, although he remained largely stuporous. As a last resort, he was re-referred for further ECT twice weekly. He again showed initial noticeable improvement with a repeat Bush-Francis Catatonia Rating Scale scored at 8. We postulate that the improved effect of ECT at this time may have been related to previous cessation of concurrent antipsychotic medication. Non-medical treatment was also offered through intensive physiotherapy, including hydrotherapy and passive stretching to reduce the risk of permanent strictures.

Despite a further 15 treatments of ECT there was no sustained benefit in psychomotor presentation. It had already been established that our patient could not tolerate high-dose benzodiazepines. By this time, the patient was confined to his water-chair, would communicate briefly verbally but he still required full nursing care for toileting, feeding, changing and transferring. He often looked anxious and the team considered medication for symptomatic relief. Sertraline was restarted to treat potential underlying depression and low-dose diazepam used for anxiety, with olanzapine added as an adjunct. There was no change in presentation one month after cessation of ECT and a best-interests decision was made with his family not to trial further treatment and to transfer him to a nursing home for full-time care.

The patient remained in a stable condition for many months in the nursing home, before sadly dying following sepsis due to bullous pemphigoid two years after his initial presentation.

**Discussion**

Catatonia is a frequently reversible condition with prompt diagnosis and treatment. Our patient was sadly refractory to treatment with benzodiazepines and ECT, and we propose several important factors in this case that may have led to the poor outcome. These include his older age, delayed recognition of catatonia, co-morbid autistic spectrum disorder and sensitivity to benzodiazepine treatment. An obvious limitation to this case report is that we were unable to identify positive prognostic factors due to the poor outcome for our patient.

Our patient’s initial presentation appeared to mimic depression, although an early clue for catatonia was increased motor activity pre-admission. Numerous potential causes for catatonia were considered and investigated including, but not limited to, severe depression, schizophrenia, dementia, limbic encephalitis, and other rarer neurological disorders. This diagnostic uncertainty led to a delay in recognition and treatment of catatonia. One small case series described delayed identification of catatonia in three elderly patients, all of whom had a poor outcome to treatment (11). We also identified our patient’s reduced speech content as a factor that precluded prompt and thorough assessment of mental state. A retrospective study examining Lorazepam treatment for catatonia showed that mutism and longer disease duration were predictors of poor outcome (17). Furthermore, a long period of untreated catatonia, and chronic catatonic states, have been associated with a poorer response to benzodiazepines (6, 18).

Abnormal baseline social interaction and response to stress meant that our patient’s previously undiagnosed autistic spectrum disorder further complicated assessment of his psychomotor signs. The delayed recognition of catatonia in autistic spectrum disorder may in part be due to overlapping clinical features between both diagnoses, including posturing, stereotypies, and alterations in motor activity (19), as shown in our patient. In young adults with autism it has been proposed that catatonia should be considered whenever there is a marked deterioration in movement, self-care and pattern of activities (20). Even if catatonia is recognised in autistic spectrum disorder, as it has been in up to 12% of individuals with autism between 17-40 years of age (20), this co-morbidity is associated with a poorer response to treatment (19). Indeed, a previous case series highlighted a very poor response to benzodiazepines in this patient group (21). An important learning point from this case report is that catatonia should be considered when there is a deterioration in presentation for anyone with autistic spectrum disorder, although it should be remembered that treatment for catatonia in this group of patients is less likely to be successful.

We believe that the age of our patient contributed significantly to several factors relating to his outcome. Catatonia is not often recognised in older adults, despite its known prevalence (22, 23) and the way in which catatonia responds to treatment in older patients is poorly understood. One case series showed that older patients were less likely to respond to ECT than younger patients (15). Older age also brings a much higher risk of adverse events when using benzodiazepines, including an increased risk of sedation, falls and fractures (24-26). There is also evidence that benzodiazepines relax the lower oesophageal sphincter and potentially make aspiration more likely (27). Our patient experienced severe adverse effects with Lorazepam and developed aspiration pneumonia when higher doses were cautiously trialled. Overall, the use of benzodiazepines is more problematic in old age, meaning that outcomes may be poorer in older individuals with catatonia.

Our patient’s long admission, which included a range of interventions in different hospital settings, prompted reflection about several ethical and practical considerations. Given the poor outcome in our patient, and the poor prognosis of treatment of catatonia in old age overall, we recommend early discussion about appropriateness of treatment options. Specifically, a shorter treatment duration may be more ethical in this age group where circumstances may indicate a much poorer prognosis. Furthermore, there are practical considerations with regards to the location of treatment provision, with intravenous Lorazepam usually only given in acute hospitals. Giving such high doses of sedating medication requires close monitoring for adverse effects and logistical practices such as nurse-to-patient ratios and monitored beds should be considered.

**Conclusions**

There remain significant limitations in our knowledge of how catatonia presents in old age. In this case report the unsuccessful treatment of catatonia in an elderly patient highlights several clinical implications for this age group. We recommend early consideration of catatonia in old age mental health services and acute medical services if relevant clinical features are present. Once catatonia has been recognised it is important to ensure the early use of ECT due to reduced tolerability of benzodiazepines in the elderly, as we know that prompt treatment improves outcomes in this age group. We recommend that further exploration of the presentation, course, and treatment of catatonia in the elderly is essential to improve clinical outcomes in this population.

**List of abbreviations**

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| CT – computed tomography |
| ECT – electroconvulsive therapy |
| GABA - gamma-aminobutyric acid |
| GP – general practitioner |
| IV – intravenous |
| MRI – magnetic resonance imaging |

**Figure legends**

Figure 1: Treatment overview during hospital admission

Timeline of treatment with electroconvulsive therapy (ECT) and medication during hospital admission, including timing of brain imaging.

**Declarations**

**Ethics approval and consent to participate:** Not applicable.

**Consent for publication:** Written informed consent was obtained from the patient’s next of kin for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

**Availability of data and materials:** Not applicable.

**Competing interests:** The authors declare that they have no competing interests.

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**Authors' contributions:** EB, CF and JA were involved in the care of the patient. EB, CF, CG and JA drafted, read and approved the manuscript.

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