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CASE REPORT

A dramatic case of a woman discovered after 7 months of untreated catatonia

M. Ratzlaff* and M. Harrington

Department of Psychiatry, University of Manitoba, Winnipeg, Manitoba, Canada

Described is an unusually severe case of catatonia in the context of a major depressive episode. The patient is a 49-year-old Caucasian female who was living with her husband in an urban apartment. In March 2015, she experienced a major depressive episode in the context of financial hardship after being dismissed from her job. She became catatonic and did not leave her apartment for 7 months. For the first 4 months she lay in bed, then after losing bowel and bladder continence, she was transferred by her husband to the bedroom floor where she lay prone for another 3 months before paramedics were notified. She subsequently underwent a 4-month admission to an intensive care unit, surgical ward, and psychiatric ward. This case shows the extreme extent of psychiatric and physical sequelae that can result from prolonged delay of treatment of severe catatonia in the context of depression.

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Description of the case

The purpose of this case report is to illustrate the extreme extent of psychiatric and physical sequelae that in rare circumstances can result from severe catatonia in the context of a major depressive episode. Although catatonia has been well described in the medical literature, no staff at our Canadian institution had ever treated or heard of a case of catatonia that had progressed to this extent of time or severity before being brought to medical attention. This case report was approved through written informed consent from the patient and review by the University of Manitoba's Health Research Ethics Board [Ethics Reference Number: HS19838 (H2016:233)].

The patient is a 49-year-old married Caucasian female who before hospitalization was living in an apartment with her husband in an urban setting. She had minimal contact with a sister who lived in the same city. She had a high school education and was unemployed, having been dismissed in March 2015 from her cashier job.

Her past medical history included a concussion, gastroesophageal reflux disease, breast reduction surgery, and a hysterectomy, all occurring years before this presentation. She was on no medications and had no known drug allergies. There was no history of alcohol or drug abuse. There was no known family history of psychiatric or neurological illness.

* Address for correspondence: M. Ratzlaff, MB, BCh, BAO, Department of Psychiatry, University of Manitoba, 771 Bannatyne Ave, Winnipeg, Manitoba, Canada R3E 3N4.
(Email: matt_ratzlaff@hotmail.com)

Regarding her psychiatric history, she had no past admissions or contact with mental health professionals. There was no history of manic or psychotic symptoms. In 2014, she had experienced one mild major depressive episode, which resolved with outpatient treatment.

After losing her job in March 2015, the patient again entered into a major depressive episode characterized by low mood, anhedonia, hopelessness, poor motivation and energy, poor concentration, memory difficulties, low appetite, and social withdrawal. For the next 7 months she did not leave her apartment. She spent increasing amounts of time lying in bed, where her husband would give her food, and eventually she remained constantly in bed except to use the toilet. She did not answer her sister's periodic phone calls, nor did she respond to her husband's requests that she seek medical help.

In August 2015, the patient completely stopped leaving her bed including for toileting. Due to her extensively soiling the bed, the husband spread a plastic sheet on the bedroom floor and relocated her onto it in a prone position. She remained conscious and lying on the floor in that position until November 2015. The husband continued to feed her on the floor and periodically cleaned up her excrement.

Eventually, the plastic sheet shifted out from under her so that the she was lying directly on the carpeted floor. She developed extensive pressure ulcers and her skin began to adhere to the carpet. Due to poor hygiene, her skin and hair became infested with maggots, lice, and flies.

In November 2015, her husband voiced concern about the patient's condition to a relative, who contacted a mobile crisis team. Upon arrival at the apartment, the team immediately called for paramedics. To move the

patient into the ambulance, paramedics had to cut the carpet off the bedroom floor as the patient's skin had adhered to it.

In the emergency department, she was found to be alert, afebrile, and vitally stable but responded to questions with only moaning. Her limbs and neck were stiff and she had severe ankle plantarflexion contractures. Due to deconditioning and contractures, she had only a flicker of movement in fingers and toes but otherwise had a normal neurological examination. She had extensive skin ulceration of her chest, abdomen, and legs. Due to the emergency department staff's concerns of neglect on the part of the patient's husband, the local police were informed, and after interviewing him, he was taken into custody.

The patient was transferred to a medical intensive care unit (ICU) for further investigation and stabilization. Due to her inability to communicate treatment wishes, her sister was made substitute decision maker. She received intravenous fluids, vasopressors, electrolyte correction, and antibiotics. Computed tomography (CT) scans confirmed skin ulcerations on the chest extending to the costal cartilages, and fluid and gas collections in the left thigh musculature and left patellofemoral joint. CT head and chest X-ray were normal.

Numerous services were consulted, including plastic surgery, orthopedics, neurology, psychiatry, wound care, medical rehabilitation, and social work. The patient underwent a series of procedures, including skin debridements, skin grafts and flaps, and vacuum dressings of her chest, thighs, and knees. At times she was able to speak words and reported ongoing low mood. An underlying psychiatric or non-psychiatric diagnosis accounting for this dramatic presentation was initially uncertain, but the Consultation-Liaison (CL) Psychiatry team and other consulted services considered depression in the differential diagnosis. However, due to the patient's inability to communicate a clear history, difficulty obtaining a reliable collateral history, and the extent of her ongoing medical comorbidities and treatments, further observation was required for clarification of a possible psychiatric diagnosis and need for psychiatric treatment.

After 11 days (late November 2015), she was transferred from the ICU to a surgical ward where she continued to be followed by the CL Psychiatry team. At this time, through further clinical observation and the limited obtainable history, a severe major depressive episode emerged as the suspected underlying diagnosis. Electroconvulsive therapy (ECT) was not felt to be indicated at this time due to the patient's unstable medical condition and ongoing surgical procedures. Antidepressant medication was commenced in late November 2015, initially with oral venlafaxine XR 37.5 mg daily and gradually titrated up to a final dose of 300 mg daily (in February 2016), which was well tolerated.

Catatonia was first suspected in late December 2015. Although no standardized catatonia rating scale was used (e.g. the Bush–Francis Catatonia Rating Scale), during bedside examinations of the patient the CL Psychiatry team began to frequently observe and elicit catatonia-related behaviors consistent with stupor (e.g. lack of psychomotor activity), staring (e.g. fixed gaze with reduced blinking), mutism (e.g. selectively not answering or using few words when questioned by staff), negativism (e.g. resisting passive arm movement, selectively closing her eyes when the treatment team was in the room), and stereotypies (e.g. purposeless fidgeting of her fingers).

Further support for catatonia was shown by dramatic, but temporary, resolution of these behaviors with lorazepam 0.5 mg orally. The first trial doses of lorazepam were given in late December 2015, and given the favorable response, several days later were increased to 0.5 mg orally scheduled three times daily plus 0.5–1 mg orally every 4–6 hours as needed. Neurology was again consulted to rule out a medical cause for her catatonia, but they concluded that it was most likely psychiatric in nature.

With a working diagnosis of a severe major depressive episode with catatonia, due to the prolonged duration of illness and limited relief with oral medications, the CL Psychiatry team felt that urgent ECT was deemed more advisable than other alternative treatments. Several attempts were made by the team, and by an ECT specialist whose second opinion was sought, to discuss with the patient the option of ECT and to obtain informed consent. However, due to her ongoing severe depressive and catatonic symptoms, she was unable to engage in this discussion and was therefore considered incompetent to provide or withhold consent. Substitute consent for ECT was obtained from the patient's sister after discussion with her of the anticipated benefits and risks and in accordance with the Mental Health Act in our Canadian jurisdiction.

Bitemporal ECT commenced in early January 2016 following a taper off of lorazepam and continued two to three times weekly for a total of 10 procedures. The mean ECT parameters were pulse width 0.7 mseconds (range 0.5–1.0 mseconds, SD 0.18 mseconds), frequency 53 Hz (range 30–60 Hz, SD 11.6 Hz), stimulus duration 6 seconds, and current 0.8 A. All treatments produced a modified grand mal seizure with mean motor duration of 27.5 seconds (range 21–37 seconds, SD 4.1 seconds) and mean electroencephalogram duration of 30.7 seconds (range 25–46 seconds, SD 6.2 seconds). Anesthesia was achieved with intravenous etomidate, propofol, or ketamine and muscle relaxation with intravenous succinylcholine.

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The patient tolerated the ECT well and did not report any side effects after treatments. Although no standardized assessment scales were used to monitor response to treatment, her clinical progress was regularly assessed through interactions with the CL Psychiatry team. After the second ECT procedure the patient was able to verbally interact with short statements and reported ongoing poor mood, energy, and concentration. By the fifth ECT procedure she was speaking in full sentences, showing an increased range of affect and increased spontaneous movement.

The patient was transferred from the surgical ward to an inpatient psychiatry ward in late January 2016 and remained there until discharge in March 2016. After the eighth ECT procedure (late January 2016), her catatonic symptoms resolved, she was showing a marked improvement in her depressive symptoms, and was thereafter able to engage in normal verbal communication with staff. It was decided to end the course of ECT after the tenth procedure, as at that time the patient was in remission from depressive symptoms and she and her sister felt she had returned to her baseline from a psychiatric standpoint.

Understandably, the patient voiced to staff feeling angry about what had happened to her before admission and also feeling frustrated about her inability to contact her husband due to an ongoing legal order. She was observed to express many questions about what happened, and why. She stated that she did know he had criminal charges relating to neglect and that he was attending court proceedings. Despite what she had endured with her husband's knowing, she consistently voiced, and submitted a videotaped statement to the courts, that she wanted all charges dropped.

A multidisciplinary, holistic approach was required to restore the patient's physical and mental health. Her prolonged immobility required daily physiotherapy to address her limb weakness and ankle contractures. She progressed from being bed-bound and totally dependent for transfers to mobilizing independently with a walker and foot braces. Urinary incontinence from pelvic floor deconditioning required sanitary care from nursing staff and eventually resolved. Her healing pressure ulcers, skin grafts and flaps required regular wound care and dressing changes.

When the patient had been adequately stabilized, a group meeting was held with the patient, her sister, and all relevant allied health professions to coordinate transition back into the community. In mid-March 2016, she was successfully discharged to live at her sister's house and continue outpatient Psychiatry and Physiotherapy follow-up. Maintenance ECT was not deemed necessary because of her complete and prolonged remission from depressive symptoms during the latter portion of her admission following her acute course of ECT, and because she was tolerating and compliant with antidepressant medication. As of June 2016, she was maintained on venalfaxine XR 300 mg daily and remained in remission from her depression. Her husband's legal proceedings regarding possible criminal charges of neglect were ongoing.

Discussion

This case is a sobering example of how catatonia can be a debilitating feature of a severe major depressive episode. It exemplifies many of the associated psychiatric and physical sequelae of catatonia, as well as the challenges in timely diagnosis and treatment. Broadly, catatonia is defined as a neuropsychiatric syndrome of motor dysregulation (either hypoactivity or hyperactivity) associated with a variety of illnesses (Wilcox & Duffy, 2014). Though historically catatonia has been most associated with schizophrenia (Tang & Duffin, 2014), modern data on psychiatric disorders show that catatonia is also commonly a feature of mood disorders (Sienaert *et al*. 2014). Furthermore, the rate of catatonia among all psychiatric inpatients may be as high as 10% (Sienaert *et al*. 2014).

As defined in the Diagnostic and Statistical Manual of Mental Disorders-fifth edition, catatonia consists of the presence of at least three of catalepsy, waxy flexibility, stupor, agitation, mutism, negativism, posturing, mannerisms, stereotypies, grimacing, echolalia, or echopraxia (American Psychiatric Association, 2013). Another commonly used diagnostic aid is the 23-item Bush–Francis Catatonia Rating Scale which, importantly, includes the staring phenomenon (Bush *et al*. 1996).

This case also illustrates how catatonia, in addition to being psychologically disabling, can be associated with a host of physical and medical sequelae requiring supportive measures. For example, the immobility of hypoactive catatonia can result in complications including pressure ulcers, contractures, deep venous thrombosis, constipation, urinary tract infections, malnutrition, and electrolyte imbalances (Coffey, 2015). As such, modern treatment of severe catatonia requires a well-coordinated biopsychosocial approach. Through collaboration of multiple medical and psychiatric departments as well as allied health professions, this patient, who due to isolation and unfortunate circumstances had developed a mental illness with physical sequelae, the severity of which is not often encountered in modern practice, was successfully restored to independence in the community.

Regarding the pharmacological treatment of catatonia in the context of depression, the Canadian Network for Mood and Anxiety Treatments (CANMAT) 2016 depression treatment guidelines only found sufficient

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(Level 3) evidence to recommend benzodiazepines (Kennedy *et al*. 2016). However, for moderate-to-severe depression in general, these guidelines also recommend as first-line (Level 1 evidence) 'second generation' antidepressants such as serotonin and noradrenaline reuptake inhibitors, selective serotonin reuptake inhibitors, agomelatine, bupropion, mirtazapine, and vortioxetine (Kennedy *et al*. 2016). The only other major North American or European depression treatment guideline directly addressing this issue is a less recent one by the American Psychiatric Association, which similarly recommends that pharmacological treatment of depression with catatonic features should generally include a combination of a benzodiazepine and antidepressant (Gelenberg *et al*. 2010).

An initial 'benzodiazepine challenge test' typically involves administration of lorazepam 1–2 mg orally or intravenously, which can rapidly but temporarily relieve catatonic symptoms (Sienaert *et al*. 2014). Thereafter, maintenance treatment may be started with lorazepam 1–2 mg every 4–12 hours and increased as needed to 8–24 mg/day. Diazepam, oxazepam, and clonazepam have also been used. Other pharmacological agents with very limited evidence of effectiveness for catatonia act on the glutamate or γ-aminobutyric acid systems and include zolpidem, amantadine, memantine, bromocriptine, biperiden, valproic acid, levetiracetam, topiramate, carbamazepine, and lithium (Sienaert *et al*. 2014).

ECT is another effective treatment for depression with catatonia, especially for severe catatonia that requires urgent intervention or does not respond adequately to benzodiazepines (Sienaert *et al*. 2014). There is a general consensus among major North American and European depression treatment guidelines that ECT is a first-line treatment for the acute phase of severe depression (with or without catatonia), especially in combination with an antidepressant medication (National Institute for Health and Care Excellence, 2009; Gelenberg *et al*. 2010; Bauer *et al*. 2013; Kennedy *et al*. 2016). For the acute phase treatment of severe depression, the CANMAT guidelines provide Level 1 evidence for first-line treatment with brief pulse right unilateral or brief pulse bifrontal ECT and for second-line treatment with brief pulse bitemporal, ultrabrief pulse right unilateral, or ultrabrief pulse bifrontal ECT (Kennedy *et al*. 2016). Bilateral ECT is more effective than unilateral, but is associated with more cognitive side effects (Kennedy *et al*. 2016). Response or remission in the acute ECT course is usually obtained after 6–15 treatments at an optimal rate of two to three treatments per week (Kennedy *et al*. 2016).

Limited evidence exists regarding maintenance pharmacotherapy or ECT treatment after remission from severe depression with or without catatonia. The CANMAT guidelines advise that antidepressant pharmacotherapy or maintenance ECT is equally effective in preventing relapse, though no studies have yet examined optimal frequency of continuation/maintenance ECT (Kennedy *et al*. 2016). If maintenance ECT is pursued, a typical schedule involves weekly treatments for 4 weeks, then biweekly treatments for 8 weeks, then monthly treatments thereafter (Kennedy *et al*. 2016).

This case also raises important ethical considerations regarding consent (and substitute consent) to ECT treatment in the context of severe mental illness. In most jurisdictions, informed consent for ECT, as with any medical procedure, is sought and obtained directly from patients when they are deemed to have capacity to do so. Guidelines for determining capacity and obtaining informed consent for ECT have been described (Mankad, 2015). However, challenges and controversies arise when patients are deemed to lack capacity to give or withhold consent to treatment. Under the Mental Health Act in our Canadian jurisdiction, and as occurred in the above case, if a patient is deemed to lack capacity to give or withhold informed consent for ECT but is suffering from a severe mental illness for which ECT treatment is felt to be in his or her best interests, then substitute consent can be sought from next of kin or other legal guardian after a discussion with them about indication and anticipated benefits and risks (Enns *et al*. 2010). A second psychiatric opinion is also strongly advised (Enns *et al*. 2010). In other jurisdictions, such as some in the United States, substitute consent for ECT treatment must usually be obtained through court approval (Dare & Rasmussen, 2015). In contrast, other jurisdictions such as Norway give primacy to patient self-determination such that ECT can be refused even in the context of severe mental illness (e.g. psychotic depression) (Moksnes, 2013). Only in life-threatening situations (e.g. persistent nutrition refusal) and where ECT is deemed to be the only adequate intervention can it be given to a non-consenting psychiatric patient, and only after court approval of a 'plea of vital necessity' from the treating clinician(s) (Moksnes, 2013). Similarly, other jurisdictions exist where if a patient is unable or unwilling to consent to ECT, substitute consent is not legally recognized. Because of these differences, it is vital that clinicians be familiar with and follow their local policies governing the delivery of ECT.

Finally, from a medicolegal perspective, this case confirms evidence that people with mental illness, including depression, are at increased risk of neglect (Dyer *et al*. 2000). The extremely disheveled state in which this patient originally arrived at the emergency department prompted staff to be concerned about neglect or abuse on the part of the patient's husband, and they acted appropriately on their duty to inform

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the local police. Clinicians are reminded of the importance of always employing a low threshold of suspicion of neglect or other abuse when assessing patients.

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Conflicts of Interest

None.

Ethical Standards

The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national and institutional committee on human experimentation with the Helsinki Declaration of 1975, as revised in 2008. The authors assert that ethical approval for publication of this case report has been provided by their local REC.

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