CASE REPORT

TREATMENT-RESISTANT CATATONIA – A CASE REPORT

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Abstract

This case report describes the clinical records of a 42 years old patient suffering from catatonic schizophrenia who was treated for approximately five months at a psychiatric intermediate care unit and subsequently for another twelve months in a specialized geriatric center. Though state-of-the-art pharmacological (antipsychotics, benzodiazepines) and non-pharmacological (electroconvulsive therapy, transcranial direct-current stimulation) treatments for catatonia were offered to the subject, no significant signs for an improvement of catatonic symptoms could be recorded using Bush-Francis-Catatonia-Rating-Scale. The case report discusses options regarding treatment-resistant psychiatric disorders. In other medical disciplines, chronic diseases often demand a palliative care setting. This might also be the case in psychiatry; however, there is a lack of methodologically sound studies on palliative care for patients with severe persistent mental illness.

Key words: catatonic schizophrenia, severe persistent mental illness, treatment-resistant, palliative care in psychiatry

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Introduction

Catatonia is a neuropsychiatric syndrome characterized by motor, behavioral, affective, cognitive and autonomic alterations (Daniels 2009). The prevalence of catatonia is unknown, presumably due to under-diagnosis by physicians as well as the fact that affected persons are partially treated in internal medicine wards, as the condition is possibly life-threatening (Häfner and Kasper 1982, van der Heijden et al. 2005). Randomized controlled trials for catatonia remain scarce (Ungvari et al. 1999) and evidence is primarily based on an extensive number of case-reports. At present, though sufficient treatments for catatonia are available, e.g. benzodiazepines, antipsychotics and electroconvulsive therapy (ECT) (Mann et al. 2004, Daniels 2009), options are limited when primary treatment approaches fail.

Case presentation

A 42 year-old man was admitted to the psychiatric intermediate care unit (PICU) (Winkler et al. 2011), following an approximately two-week period of hospitalization at a standard psychiatric care unit. The patient had been transferred due to an aggravation of the catatonic state, accompanied by incapacity for fluid and food consumption, a massive rigidity of the limbs and hyperthermia. In addition, antipsychotic treatment

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with clozapine had been paused as a neuroleptic malignant syndrome (NMS) was suspected. In the patient's medical history, obtained via the patient's brother (legal trustee), it was determined that the patient had suffered from mental retardation since childhood. Up to date no underlying organic correlate had been detected. Catatonic schizophrenia had been diagnosed at adolescence. The patient had been living in a supervised flat-sharing community and was repeatedly hospitalized after recurrent worsening of his psychiatric condition. The last catatonic episode had occurred four years earlier, however, successfully treated with ECT. Up to the current admission, the patient's psychiatric symptoms had been stable under treatment with clozapine.

During the present admission, the psychiatric state of the patient was characterized by a high degree of rigidity, tension and mutism. These symptoms resulted in a deficient capacity for self-care, demanding a treatment in accordance to the Austrian involuntary commitment law. Administration of food, fluid and medication was ensured via a nasogastric tube and a central venous catheter. At admission, the patient showed a total score of 43 (of 69) when assessed with the Bush-Francis-Catatonia-Rating-Scale (BFCRS) (Sienaert et al. 2011), with particularly high scores in excitement, immobility, mutism, staring, posturing, grimacing, rigidity and negativism. A potential somatic cause of the symptoms was excluded using magnetic resonance imaging, lumbar puncture and electroencephalography.

Upon exclusion of a NMS (rigidity and alertness instead of rigor and unconsciousness), treatment with clozapine was reinstated at a maximum dosage of 200 mg, though without success and causing hypersalivation. Following consent by the patient's trustee as well as anaesthesiologic approval, ECT was performed using a brief pulse ECT device (Thymatron System IV™). In order to achieve quick treatment response, the first treatment was administered bilaterally at a stimulus charge of 30 % (= 150 millicoulombs). Subsequently, 14 further treatments were performed up to a maximum stimulus intensity of 150 % (= 750 mC). The treatment series had to be interrupted several times due to deteriorations of the patient's somatic condition including recurrent infections (pneumonia, septicaemia) preventing more frequent and further ECT sessions. Overall, despite a slight reduction of mutism and rigidity (BFCRS = 40), ECT was not effective. Despite the anticonvulsant effect of lorazepam potentially mitigating the effectiveness of ECT, the psychiatric state of the patient did not allow for a discontinuation of benzodiazepines, which are indispensable for the treatment of catatonia (Daniels 2009). In fact, constant psychomotor abnormalities, psychotic symptoms, particularly pronounced anxiety, paranoid delusions and cenesthesia, persisted, even after consecutively switching antipsychotic treatment to aripiprazole (20 mg), risperidone (6 mg), olanzapine (20 mg) and haloperidol (10 mg) (see table). As lorazepam (daily dose 16 mg) showed an insufficient sedative and relaxing effect, and as related autonomic abnormalities, especially hypertonia, tachycardia, hyperthermia and intensive transpiration persisted, a midazolam infusion pump (3-5 mg/h) was implemented.

Subsequently, the patient had to be transferred to an internal intensive care unit (ICU), where he was intubated, mechanically ventilated and as a consequence a tracheostomy was accomplished due to pneumonia and resulting sepsis. Contrary to expectations, sedo-analgesia (see table) did not reduce the patient's tension. While weaning, the slow withdrawal of sedo-analgesia resulted in a recurrence of catatonia.

Back at PICU, a PEG (percutaneous endoscopic gastronomy) tube was inserted in order to exclude the nasogastric tube and the central venous catheter as possible sources of infection. Medication was administered via the PEG tube. Based on several positive and promising case-studies in treatment-resistant schizophrenia, catatonic schizophrenia as well as persistent hallucinations (Shiozawa et al. 2013) transcranial direct-current stimulation (tDCS) was performed daily for a period of ten consecutive days during 20 min with a stimulus intensity of 2 mA (DC-stimulator, neuroConn, Germany) upon obtainment of the trustee's consent. However, the mental and psychomotor state of the patient remained nearly unchanged (BFCRS = 37). Finally, after 147 days of hospitalization, there was general agreement that no further treatment options were available for this patient at our ward and therefore he was transferred under psychopharmacological treatment with daily doses of diazepam 65 mg, haloperidol 5 mg and olanzapine 30 mg (see table) to a specialized geriatric center where he would hopefully benefit from further intensive nursing care with the option of long-term ventilation. One year after discharge, the patient was still in an unchanged condition.

Discussion

Case-reports in which state-of-the-art pharmacological and non-pharmacological approaches are unsuccessful are rare. In clinical practice, however, discussion regarding options for treatment resistant patients is essential. In other medical disciplines, for example in oncology, severe, chronic diseases often demand a palliative care setting when no cure can be expected. The goal is to improve quality of life with a focus on stress and pain relief. This might also apply in psychiatry, particularly when somatic health is at risk. Furthermore, chronically mentally ill persons exhibit an increased mortality (Brown and Birthwhistle 1996), e.g., in schizophrenia, severe anorexia nervosa or borderline personality disorder with recurrent, dramatic self-injuries. In contrast to somatic disorders, the prognosis of a life-threatening mental illness cannot be derived from objective and measurable markers, apart from those of related somatic comorbidities. Interestingly, this issue is mainly addressed in the scientific nursing literature (Baker 2005, Terpstra and Terpstra 2012). The term of *palliative care psychiatry* can be found in the literature; however, these articles refer primarily to psychiatric syndromes commonly experienced by patients in palliative care settings suffering from incurable somatic disorders. According to a recent review, 50% of palliative care patients suffer from depressive symptoms, 70% of anxiety and nearly all from delirium as death nears (Fairman and Irwin 2013).

It is a topic of discussion whether psychiatrists are ethically in a position to decide whether natural death should be allowed for a person with persistent catatonia. The controversial term of medical futility has been frequently used in the literature in the context of treatment-resistant anorexia nervosa and heroin-addiction questioning unconditional, compulsory therapy. Case-reports of patients ultimately referred to palliative and hospice care settings following years of unsuccessful treatments were published repeatedly (Lopez et al. 2010). A comprehensive literature review published recently concludes that continuous psychiatric treatment of a severe disorder can never be considered as futile (Geppert 2015). However, as yet there is no consensus on best practice or a specific therapy setting for these patients, but a palliative approach might represent a beneficial option

Summary of different treatment options applied to the catatonic patient during the hospitalization period of 147 days in chronological order. PICU, psychiatric intermediate care unit; ICU, internal medicine intensive care unit; ECT, electroconvulsive therapy (Thymatron System IV™); tDCS, transcranial direct-current stimulation, (DC-stimulator, neuroConn, Germany). Psychotropic maximum daily doses (DD). are given in mg; maximum stimulus intensity of ECT and tDCS in mC and mA, respectively

Ward	Treatment	Week 1	Week 2	Week 3	Week 4	Week 5	Week 6	Week 7	Week 8	Week 9	Week 10	Week 11
PICU	antipsychotics (DD)	clozapine 200 mg	clozapine 200 mg	clozapine 200 mg	no treatment	no treatment	clozapine 100 mg	aripiprazole 15 mg	olanzapine 20 mg	olanzapine 20 mg		
	sedatives (DD)	lorazepam 8 mg	lorazepam 8 mg	lorazepam 8 mg	lorazepam 8 mg	lorazepam 8 mg	lorazepam 8 mg	lorazepam 8 mg	haloperidol 10 mg	haloperidol 10 mg		
	non-pharmacological	bifrontal ECT (300 mC)	bifrontal ECT (400 mC)	bifrontal ECT (500 mC)	bifrontal ECT (500 mC)	bifrontal ECT (500 mC)	bifrontal ECT (500 mC)	bifrontal ECT (650 mC)	midazolam 3-5 mg/h	midazolam 3-5 mg/h		
									-	-		
Ward	**Treatment**	**Week 1**	**Week 2**	**Week 3**								
ICU	antipsychotics (DD)	olanzapine 20 mg	olanzapine 20 mg	olanzapine 20 mg								
	sedatives	haloperidol 10 mg	haloperidol 10 mg	haloperidol 10 mg								
		midazolam 5-10 mg/h	-	midazolam 3-5 mg/h								
	others	propofol 160 mg/h	propofol 100 mg/h	-								
		remifentanil 0.8 mg/h	remifentanil 0.2 mg/h	remifentanil 0,1 mg/h								
Ward	**Treatment**	**Week 1**	**Week 2**	**Week 3**	**Week 4**	**Week 5**	**Week 6**	**Week 7**	**Week 8**	**Week 9**	**Week 10**	**Week 11**
PICU	antipsychotics (DD)	olanzapine 20 mg	olanzapine 30 mg	olanzapine 30 mg	olanzapine 30 mg	olanzapine 30 mg	olanzapine 30 mg	olanzapine 30 mg	olanzapine 30 mg	olanzapine 30 mg	olanzapine 30 mg	olanzapine 30 mg
		haloperidol 10 mg	haloperidol 10 mg	haloperidol 10 mg	haloperidol 10 mg	haloperidol 10 mg	risperidone 3 mg	risperidone 6 mg	risperidone 6 mg	risperidone 6 mg	haloperidol 10 mg	haloperidol 10 mg
	sedatives (DD)	lorazepam 6 mg	lorazepam 6.75 mg	lorazepam 6.75 mg	lorazepam 6.75 mg	lorazepam 6.75 mg	lorazepam 6.75 mg	diazepam 70 mg	diazepam 70 mg	diazepam 70 mg	diazepam 70 mg	diazepam 70 mg
	non-pharmacological	-	-	-	-	-	-	tDCS (2 mA)	tDCS (2 mA)	tDCS (2 mA)	-	-

Time →

for some of the affected individuals, including the patient described in the present report (Trachsel et al. 2016). Due to the lack of prognostic criteria and consistent guidelines for treatment resistance, at present, the therapy of severe cases of persistent psychiatric disorders has to be discussed individually having regard to the legal trustee's input (Ruggeri et al. 2000). We offered intensive-care management and subsequently care at a geriatric center with the possibility for long-term ventilation. The lack of methodologically sound studies on palliative care for patients with severe persistent mental illness and the need for the development of a new treatment paradigm for these disorders was addressed in the literature (Woods et al. 2008, Trachsel et al. 2016); however, up to date little has changed. The present case-report is a clinical example highlighting the need for further investigations that help to establish treatment guidelines for currently untreatable psychiatric conditions.

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