Electroconvulsive therapy in a case of catatonia with severe somatic complications

Trattamento con terapia elettroconvulsivante di un caso di catatonia complicato da gravi manifestazioni somatiche

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Summary

Background
Catatonia is a neuropsychiatric syndrome that may occur in association with mental, neurological and medical disorders. A delay in diagnosis and treatment of catatonic symptoms is related to a high risk of medical complications such as dehydration, malnutrition, pressure ulcers, thrombotic events, aspiration pneumonia and infections.

Objectives
The authors present the case of a bipolar patient, admitted to the Psychiatric Clinic of the Azienda Ospedaliero-Universitaria Pisana for catatonic syndrome, complicated by weight loss, deep vein thrombosis (DVT), pressure ulcers and systemic infection.

Results
Supportive therapy, including hydration, electrolytic restoration and antibiotics was adopted to stabilize the patient’s general conditions. Treatment with low molecular weight heparin was given for DVT and to prevent pulmonary embolism. Catatonic symptoms were initially treated with intravenous administration of delorazepam, with some improvement in catalepsy and waxy flexibility. As treatment with benzodiazepines was not completely effective, electroconvulsive therapy (ECT) was used in combination with delorazepam, which led to progressive resolution of catatonic symptoms.

Conclusions
The existence of medical complications requires a multidisciplinary therapeutic strategy, with the intervention of different specialists. Our experience confirms the efficacy and safety of ECT in catatonia, even in the presence of serious complications such as DVT. In these cases, combination of ECT and benzodiazepines should be considered elective treatment to achieve quick resolution of symptoms and reduce morbidity and mortality.

Key words
Catatonia • Bipolar disorder • Electroconvulsive therapy • Benzodiazepines

Introduction
Catatonia is a complex neuropsychiatric syndrome characterized by onset of psychomotor manifestations (negativism, mutism, mannerisms, stereotypies, immobility, rigidity) in association with a number of pathologies, not only psychiatric, but also physical such as neurologic, toxicologic, endocrinologic or infective. Despite the original description of catatonia by Kalbahum 1 , who highlighted some clinical characteristics such as cyclic course, interepisode remission and frequent association with affective symptoms shared with mood disorders, for many years, influenced by the studies of Kraepelin and Bleuler 2,3 , catatonia has been classified as a subtype of schizophrenia. In recent years, several clinical and epidemiological studies 4-6 have shown that catatonia is not only underdiagnosed, but that patients with bipolar disorder have a higher prevalence of catatonic symptoms than patients with schizophrenia. Thus, both clinicians and researchers have acknowledged the need for better classification and diagnosis. Other authors, such as Taylor and Fink 7 , have proposed that the next edition of the DSM should classify catatonia as a separate diagnostic entity, while still others have stressed the need to reformulate diagnostic criteria, noting the currently inadequate psychopathologic description and the lack of evaluation criteria on severity and duration 8-9.

The main epidemiological studies 10 , even if conducted in apparently homogenous patient populations, such as psychiatric in-patients, report a prevalence of catatonia that varies widely, from 7% to 31%. Clinical expression of catatonia, in fact, includes a large and variable spectrum of motor, behavioural, affective and cognitive manifestations (Table 1) that consider the complexity of presentation and diagnostic difficulties. Mutism, negativism and catalepsy are classic symptoms that are...
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Antagonists may have a role in treatment of catatonia. Two case reports have also described the use of repeated transcranial magnetic stimulation (TMS) while the role of atypical antipsychotics is still under investigation.

Clinical case

D. is a 43-year-old woman, divorced, with a 24-year-old daughter. She had been previously followed by Psychiatric Services at a hospital near her home for bipolar type I disorder with psychotic features, and was admitted to our Psychiatric Clinic following the appearance of catatonic manifestations. Clinical history, with the help of family members, revealed the onset of psychiatric symptoms following puerperium, with a depressive episode having both mood-incongruent and mood-congruent psychotic features. In later years, the patient presented numerous recurrences with the same symptoms that needed hosp-
The patient showed gradual sitophobia (she suspected poisoning) that led to progressive, severe weight loss (about 35 kg in 3 months), with hospitalization at a local psychiatric clinic. Despite a number of therapies, mostly typical and atypical antipsychotics (haloperidol, olanzapine, risperidone, clozapine) and benzodiazepines (delorazepam), there was no improvement in clinical symptoms, and catatonic manifestations began to develop (psychomotor arrest, mutism, negativism, rigidity with catalepsy, sitophobia, oppositionity). The patient was transferred to our clinic, which is a level III regional centre for severe and complex pathologies, and is licensed to perform ECT.

Upon admission, the patient was under treatment with risperidone (1.5 mg/day), delorazepam (2 mg/day) and enoxaparin sodium (4000 IU/day, s.c.), was immobile, stuporous, with waxy flexibility. She was mute, negativist and oppositional and refused both food and therapy. Severe malnutrition was present (BMI = 15 kg/m²), with marked diffuse muscular hypotrophy and dehydration. Objective examination revealed a deep vein thrombosis (DVT) at the popliteal region of the left leg, grade IV pressure ulcer at the sacral level and modest fever (37.8°C). Arterial pressure, cardiac frequency and ECG were within normal limits. Laboratory investigations showed no relevant alterations with the exception of modest hypoalbuminaemia (3.14 mg/dl) and high levels of D-dimer (0.67 mg/dl). An emergency encephalic CT was carried out with negative results, while objective neurological examination revealed waxy flexibility in all four limbs that was more predominant in the arms, an a distal tremor at resting in the left arm, which could be attributed to iatrogenic damage subsequent to neuroleptic therapy. Therapy with delorazepam (8 mg/day i.v.) and systemic antibiotics were initiated (levofloxacin and teicoplanin i.v.) for pressure ulcers at the sacral level. A urinary catheter and a nasogastric tube for parenteral nutrition enteral were inserted after placing a peripherally inserted central catheter (PICC). Antiplatelet therapy with enoxaparin was administered, and it was also decided to start topical therapy for the pressure ulcers with collagenase and polyurethane film.

On the first day after admission, the patient initiated diagnostic work-up for ECT. Cardiac and respiratory pathologies that would contraindicate ECT were excluded by echocardiography and thoracic radiography. Echo colour Doppler revealed the presence of endoluminal thrombotic material involving the left superficial vein, popliteal vein and several of the homolateral subpopliteal veins. To avoid pulmonary embolism, in anticipation of ECT, antiplatelet therapy was substituted with dalteparin sodium 10,000 IU/day.

In the following days, while the psychopathological picture was substantially unvaried, except for the loss of waxy flexibility, the general conditions of the patient began to worsen with an increase in febrile episodes (> 38°C) until reaching a state of constant hyperpyrexia (with the appearance of frequent bigeminal ventricular extrasystoles by ECG), in spite of a change in antimycotic i.v. therapy (tigecycline, fluconazole) based on the results of the antibiogram of cultures from the pressure ulcer (S. haemoliticus, E. coli, C. albicans). Haemoculture was positive for Pseudomonas aeruginosa. Considering this, at 14 days after admission, the patient was transferred to the Internal Medicine department, and three days later, to the Intensive Care Unit due to loss of ventilatory capacity. In the ICU, vital functions were stabilized and after a negative cerebrospinal fluid test, with consent of her legal guardian, began a cycle of ECT. Four applications of ECT were performed during the first week in the ICU. Therapy with dalteparin sodium was effective in prevention of pulmonary embolism: vital signs remained stable, peaks of fever were reduced in both frequency and entity (< 38°C), although anaemia was revealed (Hb 8 g/dl), which was likely not identified at admission due to haemoconcentration. The psychopathological state was not substantially modified, and catatonic manifestations continued. On day 28 after admission, considering the improvement in general somatic conditions, it was decided to transfer the patient back the Psychiatric Clinic. Enteral nutrition was supplemented with parenteral nutrition, and system antibiotic treatment was continued (meropenem, amphotericin B), as well as antithrombotic therapy with dalteparin sodium s.c. in addition to i.v. delorazepam. At 32 days, a fifth application of ECT was administered, and the patient began to show signs of reawakening: she asked who the doctors were, where she was and why she was in hospital. She responded adequately to questions, and showed preservation of memory of past events, even if there was some amnesia about events in recent months. The patient was disoriented from a spatial-temporal standpoint, but recognized people and familial objects. Passive mobility to all four limbs was normalized, while active mobility was present but limited to severe and diffuse muscular hypotrophy. Upon interview, neither delusional thinking nor dysperceptions were present, while assessment of the affective domain showed a mild depressive symptoms that were partly due to pain from the pressure ulcer. The next day the patient began to feed herself, and enteral and parenteral nutrition were substituted with a semiliquid diet integrated with protein and potassium.

During the next 10 days, the patient received another 5 ECT sessions, with consolidation of previous results. Dur-
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Here, we present a case of a patient with bipolar I disorder, with post-partum onset at the age of 20 years, accompanied by a depressive psychiatric episode. In the following years, the disorder was characterized by recurrent affective episodes of prevalent mixed/psychotic polarity, with partial recovery and persistence of psychotic symptoms in the intervals between episodes. The last episode, occurring 3 months before the patient came under our observation, needed recovery at a psychiatric clinic near the patient’s home for severe weight loss due to si-tophobia, sustained by fear of poisoning. The successive worsening of clinical conditions, with the appearance of catatonic manifestations such as mutism, negativism and rigidity, led to her transfer to our clinic.

Correct diagnosis required identification of the pathology at the basis of her condition, together with the recognition of catatonic symptoms. Even in the presence of a positive psychiatric anamnesis, such as in the present case, it is necessary to exclude that catatonic symptoms have a somatic origin, taking particular attention to acute, life-threatening pathologies such as infective, neurologi-cal or toxicological (meningoencephalitis, stroke, cerebral haematoma, brain tumour) 12. Thus, in the first instance, objective psychiatric assessment must be integrated with accurate general and neurological examination; based on these findings, additional laboratory and instrumental investigations can be carried out.

In the case described, laboratory exams and encephalic CT upon admission excluded a somatic cause, and confirmed a clinical diagnosis of a mixed episode of bipolar type I disorder with catatonic manifestations. In anticipa-tion of ECT, the patient underwent additional diagnostic work-up that included cardiologic, neurologic and other investigations (encephalic CT, ECG, thoracic radiography) to exclude contraindications for ECT. The prolonged immobility and malnutrition led to the appearance of severe complications including weight loss, diffuse muscular hypotrophy, dehydrations, DVT in the popliteal region and grade 4 pressure ulcers at the sacral level. Thus, a urinary catheter was promptly inserted along with a nasogastric tube for enteral nutrition, later integrated with parenteral nutrition following the placement of a PICC. The early administration of antibiotic prophylaxis (pulmonary, urinary, pressure ulcers) did not prevent progression of infection for which the patient was transferred first to Internal Medicine and then to the ICU. The patient’s recovery in the ICU was decisive for resolution of sepsis and stabilization of vital signs. Our experience also confirms the crucial role of an early and integrated multidisciplinary approach to a catatonic patient involving psychiatrists, internists, infectologists, anaesthesiologists and physiotherapy specialists. In expectation of ECT, particular attention was paid to the risk of pulmonary embolism associated with DVT at the popliteal area of the left leg.

In the literature, pulmonary embolism is one of the most feared complications that an untreated catatonic patient
can encounter. The increased risk for thromboembolic events is mostly related to immobility, although the concomitant presence of pre-existing cardiovascular comorbidities or other complications such as dehydration are also factors for increased risk. Further information is needed, however, on the association between ECT and the risk of thromboembolic events as only rare cases of pulmonary embolism have been described in the literature in patients subjected to ECT. In reality, a direct cause-effect relationship between ECT and thromboembolic events has not been demonstrated, although the efficacious use of ECT in catatonic patients with a recent history of pulmonary embolism and anticoagulant treatment has been described.

For prophylaxis of thromboembolic events, our patient was administered enoxaparin (4000 IU/day s.c.) which was substituted with dalteparin sodium (10,000 IU/day s.c.). This strategy was effective even during ECT, confirming the validity of indications in the literature regarding the use of low molecular weight heparin formulations for prevention of thromboembolic events in catatonic patients.

On the basis of literature data, treatment of catatonia with整顿al administration of benzodiazepines and ECT should be considered elective. Lorazepam is the most frequently used drug, with a reported remission rate of 70% and 80%. In Italy at present, lorazepam is not available in a liquid formulation, which is a serious limitation in management of catatonic patients; in the case described, we administered a drug with a high efficacy, namely delorazepam (8 mg/day i.v.). Considering the reported synergism between ECT and benzodiazepines, some authors have suggested that combined treatment with ECT and lorazepam should be initiated immediately, especially when the critical conditions of the patient require rapid resolution of catatonic symptoms.

In the present case, initial treatment with i.v. delorazepam led to only a slight improvement in neurologic symptoms; therefore, once vital signs were stabilized, a first cycle of 10 sessions of ECT over 2 weeks was initiated, which led to a progressive improvement in both affective and motor symptoms. In literature, it is reported that, especially in severe cases of malignant catatonia at initial management phase, several applications of ECT should be administered over short intervals, sometimes with daily sessions.

After remission of catatonic symptoms in our patient, depressive symptoms with psychotic features appeared, which was resistant to initial treatment with mirtazapine and olanzapine. It was therefore decided to initiate a second cycle of 6 sessions of ECT over a longer time interval of one month. This led to significant improvement of depressive symptoms and reinforcement in the improvement of catatonic symptoms. The patient was discharged in good clinical conditions with the ability to feed herself and walk autonomously, despite of the difficulties due to muscular hypotrophy, and in a stable psychopathological state, characterized by residual psychotic symptoms such as self-reference and interpretive schemes.

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References

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