

Expanding the Catatonia Tent

Recognizing Electroconvulsive Therapy Responsive Syndromes

Max Fink, MD

Abstract: Catatonia is a motor and mood disorder of behavior increasingly recognized in systemic medically ill. Neuroleptic malignant syndrome, delirious mania, self-injurious behaviors in autism, and limbic encephalitis are conditions in which ECT-responsive catatonia is increasingly recognized and effectively treated.

Key Words: catatonia; malignant catatonia; neuroleptic malignant syndrome; delirious mania; self-injurious behaviors; autism; limbic encephalitis; benzodiazepines; lorazepam challenge test; electroconvulsive therapy; schizophrenia

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Catatonia is an acute onset systemic behavior syndrome with prominent motor and mood features that is increasingly recognized from 3% to 15% of hospital inpatients, usually in emergency, medical, neurology, and psychiatry units. Stupor, mutism, negativism, rigidity, posturing, staring, and 20 other behaviors are the classical signs described for more than a century. When first delineated in 1874 by the German psychiatrist Karl Kahlbaum, he noted "... the obvious association of this illness with other signs of disease, and its constant occurrence with certain somatic (particular muscular) disorders have been more or less ignored." Soon recognized by others, Emil Kraepelin placed his catatonia cases within his concept of dementia praecox in 1899. A decade later, the Swiss psychiatrist Eugen Bleuler reimagined the illness as *schizophrenia*, with catatonia as a marker of the illness. It remained so incorporated until it was exhumed in the 1990s with the development of defining rating scales, verification tests, and effective treatments, bringing catatonia within systemic medicine praxis.^{1,2}

Many behaviors are being drawn into the catatonia tent with four described here—neuroleptic malignant syndrome, delirious mania, self-injurious behavior (SIB) in autism, and limbic encephalitis. Recognizing catatonia assures effective, often lifesaving, treatments; first with benzodiazepines, and when these fail, with electroconvulsive therapy (ECT).

By the 1970's academic psychiatry developed rating scales to measure the presence and severity of abnormal behaviors. Research Diagnostic Criteria proposed by the clinicians at Washington University offered measures for 14 disordered behaviors. A 1973 review of 2500 file records of hospitalized mentally ill at the University of Iowa identified 10% as exhibiting signs of catatonia. Follow-up records showed that 40% had responded to medication and electroshock therapies, leading the investigator to opine that these surely had not been ill with schizophrenia, which has a much lower recovery rate.³

From the Departments of Psychiatry and Neurology, Stony Brook University School of Medicine, Stony Brook, NY.

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Reprints: Max Fink, MD, Stony Brook University School of Medicine,

631.637.1730, 11 Buttonfield Lane, South Hadley, MA 01075

(e-mail: maxfink55@gmail.com).

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A 1976 survey of admissions to the psychiatric service of a municipal New York City hospital found 55 cases of catatonia, with only 4 meeting the schizophrenia criteria, with 2 of 3 affectively ill, the majority meeting mania criteria.⁴ Also, a report of 8 patients with catatonia induced by treatment with high-potency neuroleptic drugs additionally questioned catatonia as a specific marker of schizophrenia.⁵

NEUROLEPTIC MALIGNANT SYNDROME

To these findings, catatonia had been erroneously visualized as a form of schizophrenia, and a new question arose with increasingly frequent reports of toxicity to potent neuroleptic drugs. In 1960, French authors recognized altered consciousness, motor rigidity, fever, and autonomic instability after the administration of haloperidol as *syndrom malin*. Year by year, additional case reports described toxic responses to potent neuroleptics. By 1980, a review of 60 neurotoxic cases specified the neuroleptic malignant syndrome (NMS), a name that was widely adopted.⁶ Dopamine receptor downregulation was considered the cause; dopamine agonists, such as bromocriptine, were the recommended treatment. Fever in the syndrome suggested a pathophysiological connection with malignant hyperthermia, for which the antispasticity drug dantrolene was recommended.

As interest in catatonia had been heightened by its separation from schizophrenia, clinicians noted the frequent appearance of mutism, rigidity, negativism, and posturing in NMS and asked whether these were signs of catatonia.⁷ Treatment trials with barbiturates reduced symptoms in half the cases, confirming catatonia.⁸ As amobarbital was being replaced in clinical practice by benzodiazepines, successful trials with lorazepam^{9,10} and diazepam¹¹ confirmed the connection of NMS to catatonia. Two retrospective reviews of NMS case material assessing scores on catatonia scales reported that 24 of 27 cases and 21 of 22 cases of NMS met catatonia criteria.^{12,13}

Clinicians next turned to ECT, finding relief of catatonia within 3 to 5 days. Confirming NMS as a treatable form of catatonia strengthened the awareness that catatonia was not a form of schizophrenia, assuring today's understanding as an identifiable syndrome with effective treatments.¹⁴

DELIRIOUS MANIA

Delirious mania, a syndrome of acute onset of excitement, grandiosity, emotional lability, delusions, and insomnia characteristic of mania, and the disorientation and altered consciousness characteristic of delirium, is another syndrome recently brought into the catatonia tent. Such patients are excited and hyperactive, talk loudly, act wildly, and grandiose. They are fearful, hide in closets, or run wildly and naked in the streets. Sleep is poor, feeding rejected, dehydration frequent. Consciousness fluctuates rapidly from excitement to somnolence, speech is slurred, orientation and memory impaired. Manic behavior, lethargy, mutism, posturing, and staring are interspersed. Brought to emergency rooms, often by police as the excitement has become uncontrollable, injected neuroleptics inhibit action but often precipitate the febrile neuroleptic malignant syndrome.

Credit is given to Luther Bell at Boston's McLean Hospital in 1849 for the early descriptions of 40 cases of mania and fevers with 3 of 4 being fatal. Reports of similar cases from among manic

patients occasionally dot the literature with most treated with lithium and neuroleptics. Finding the response to these therapies to be slow, with frequent precipitation of febrile states, encouraged attention to the signs of catatonia and its treatments.

As clinicians assured themselves that catatonia was not schizophrenia, they identified prominent catatonia signs in patients admitted in delirious manic states, usually in restraints and maintained on the ward under 24-hour observation. Recognition of malignant catatonia by Arnold and Stepan¹⁵ in 1952 encouraged invoking the daily induction of seizures to assure rapid and effective relief. Treatments with ECT resolve the excitement in 2 to 3 days and clarify the delirium within a week, especially when seizures are induced daily.¹⁶ Numerous reports of similar experiences dot the literature, the most recent verifying the lifesaving nature of recognizing and treating the syndrome.¹⁷

LIMBIC ENCEPHALITIS

Limbic encephalitis, an acute neurological disorder, was first described in the 1960s as a “paraneoplastic condition”—one resulting from tissue changes induced by tumors that trigger a form of self-poisoning. It is considered an autoimmune disorder, a condition in which the immune system mistakenly attacks and destroys healthy body tissue. More than 80 different autoimmune disorders are described. The pathophysiology is poorly understood, and the empiric treatments depend on corticosteroids and immunomodulatory therapies.

An “anti-NMDAR encephalitis” reaction is present in the serum and cerebrospinal fluid of patients who develop an acute multistage illness that progresses from psychosis, memory deficits, seizures, and language disintegration into a state of unresponsiveness. Patients have early-onset headache, fever, nausea, vomiting, diarrhea, or upper respiratory symptoms. Behavior is altered with the catatonia signs of mutism, negativism, and echopraxia. A report of 100 cases of this syndrome exquisitely describes the signs of catatonia in the patients. Dalmau et al,¹⁸ the originator of the NMDAR test, cites catatonia as a prominent feature of the illness.

A possible connection to tumors sends patients to whole-body searches, but the syndrome is also accepted as a diagnosis without finding a tumor. In normal life, various tissue receptors, one of which is the *N*-methyl-D-aspartate receptor, are part of the body's natural physiology. In a combined series of 577 patients, tumor removal in 357 patients relieved 220 (61%); immunotherapy relieved 53%. Overall, these treatments are slow in their response.

Persistent psychiatric symptoms and failures of psychotropic drugs sends patients to ECT. In the latest review of his experience, Dalmau et al¹⁹ described 30 cases treated with ECT with 65% reported improved. He noted that 21 had been treated before an awareness of a positive test.

Considering the presence of catatonia among patients with NMDAR encephalitis offers a successful course of treatment, with both benzodiazepines and ECT.²⁰

SELF-INJURIOUS BEHAVIORS IN AUTISM

Child psychiatrists treating autistic children with self-injurious behaviors recognized the posturing, rigidities, repetitive acts and shrieks, and refusals of feeding and toileting as signs of catatonia. Desperate for relief, trials with ECT remarkably relieved the repetitive self-injurious behaviors (SIB).

Over the next decade, 22 long-term hospitalized severely autistic adolescent patients with incapacitating self-injurious behaviors were treated with ECT at Baltimore's Kennedy Krieger Institute. Six girls, 16 boys aged between 8 and 26 years with prominent signs of catatonia were first unsuccessfully treated with benzodiazepines. At the time of the report, the patients had received from 16 to 688 ECT

with 13 on courses of continuation ECT. Negativism, SIB, and other motor behaviors resolved sufficiently for the patients to return to their homes and community schooling. The behaviors were modified but continuation of ECT at home settings were necessary.²¹ An accompanying report detailed the signs of catatonia finding stereotypics, posturing, negativism, mutism, and stupor most common.²² Video records of patients ill and after treatment are convincing that SIB in autism is accompanied by numerous signs of catatonia that are responsive to ECT.

The patients in these studies are among the more severely ill in the autism community. But many children with less severe illnesses are repetitive in speech and acts, negativistic refusing food, posturing, and mute. Considering catatonia offers alternate avenues for a treatable diagnosis.

Treatment Algorithms Are Successful

Recognizing catatonia offers means for rapid resolution. Identification during examination is guided by signs cited on a catatonia rating scale. The presence of two or more signs is sufficient to warrant further examination. Verification of the diagnosis is made by the relief to an intravenous dose of lorazepam (1–2 mg). A reduction in signs of 50% or greater is immediate verification. Such tests verify the presence of catatonia in 70% of case reports.²³

Successful treatment by 0.5 g intravenous amobarbital was first reported in 1930. It was quickly established as the emergency treatment of mutism and negativism. When benzodiazepines were recommended replacements for barbiturates, diazepam, and lorazepam became the effective treatments. Dosages must be high (6–20 mg lorazepam daily). In some countries, zolpidem is the standard treatment.

Treatments are effective when proper dosages are administered. In hospitalized patients with stupors, 70% of patients respond fully to high-dose benzodiazepine treatment. Electroconvulsive therapy is the effective treatment for the rest; it is the primary treatment in delirious mania, malignant catatonia, self-injurious behaviors in adolescents, and acutely ill limbic encephalitis. Among the variations in technical aspects of ECT, the routine use of bilateral electrode placement, high energy levels determined by age, and daily induced seizures are accepted for these seriously ill.²⁴

The Biology of Catatonia

How are we to envision catatonia among the systemic illnesses that make up the body of medicine? Catatonia is a disorder of posture, movement and speech, with many patients reporting intense anxiety and fear. It does not result from a structural defect in a single body organ nor is it identified with a physiologic dysfunction. It is not the consequence of a structural brain lesion. It occurs widely as a feature of systemic medical illnesses. After catatonia is relieved we see no residuals; it is as if the blackboard has been erased with a few smudges left at the corners. It is a behavior of the whole organism, arising suddenly and vanishing without a trace. It is likened to the inherited behaviors of sleeping, crying, coughing, or sneezing, fevers associated with systemic infections, or patterned grand mal seizures.²⁵

Catatonia is distinct from other behaviors defined as psychiatric illnesses. It is not a disorder in thought or emotion, although such accompaniments are common, particularly melancholia and mania. Some authors consider catatonia as an ‘end-state’ whole-body response to imminent doom, a behavior inherited from ancestral encounters with carnivores, an adaptation that remains an inherent feature of living. This would make catatonia an atavism.²⁶

Kahlbaum described his patients as “*astonished*” or “*thunderstruck*.” Catatonia appeared “*after very severe physical or mental stress . . . such as a terrifying experience*”; “*the patient remains motionless, without speaking, and with a rigid masklike*

facies, the eyes focused at a distance . . . devoid of any will to move or to react to any stimulus". He continues: "The general impression conveyed . . . is one of profound mental anguish, or an immobility induced by severe mental shock". Citing fear as the central theme of the syndrome, Kahlbaum titled his book *Catatonia, The Tension Insanity*.²⁷

L'Envoi

Catatonia is a recognizable, verifiable, and treatable behavior syndrome with many guises. The first signs characterized as catatonia were mutism, negativism, posturing, and stupor. Also recognized were periodic excitement. All entities identified so far are responsive to the known interventions; indeed, it is often their responsiveness that assures their recognition.

Other conditions with prominent catatonia features successfully treated by ECT are melancholia, Tourette's Syndrome, and obsessive-compulsive disorders, each of which exhibits catatonia features responsive to catatonia treatments, including ECT.²⁸ The motor aspects of melancholia and the prevalence of catatonia signs were detailed by Parker and Hadzi-Pavlovic in 1996²⁹ and by Shorter and Fink in 2010.³⁰

The exhumation from its century-long burial in schizophrenia warrants catatonia being brought into the body of clinical medicine, rather than its present place in clinical psychiatry. Its development into an identifiable, verifiable, and treatable systemic syndrome is an unheralded achievement in medical history.

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