Catatonia: The Ultimate yet Treatable Motor Reaction to Fear in Autism
Dirk M Dhossche
Department of Psychiatry, University of Mississippi Medical Center, Jackson, Mississippi, USA

Abstract

Background: Catatonia is a unique syndrome characterized by specific motor signs, at times life-threatening when aggravated by autonomic dysfunction and fever, yet treatable if recognized early. Catatonia occurs in patients with various disorders including autism and related developmental disorders. Sometimes catatonia develops after severe psychological trauma, supporting the view that it is an extreme motor reaction to fear.


Results: There are no empirical studies in the literature addressing the role of fear in the development of catatonia in autism. Case-reports support that stressful events often precede the development of catatonia in autistic people and that catatonia in autism is a treatable syndrome, often requiring psychosocial interventions to reduce anxiety, and the use of benzodiazepines, electroconvulsive therapy, and maintenance electroconvulsive therapy for safe reversal. Autistic people may be particularly vulnerable to catatonia due to social, cognitive, and sensory deficits.

Conclusion: Further studies are warranted in autistic people, measuring states of anxiety in response to various stressors, and assessing their relationship to catatonia, and applying various treatments including benzodiazepines, electroconvulsive therapy, and psychosocial interventions, in those with catatonia.

Keywords: Catatonia; Autism; Fear; Anxiety; Motor; Tonic; Immobility

Introduction

Catatonia is a unique syndrome characterized by specific motor signs, at times life-threatening when aggravated by autonomic dysfunction and fever, yet treatable with benzodiazepines and electroconvulsive therapy (ECT) if recognized early [1,2].

Signs observed in catatonia include immobility or severe motor slowing, sometimes alternating with excessive motor activity that is mostly purposeless and not influenced by external stimuli, extreme negativism, reduction of speech or mutism, repetitive movements or stereotypes, echolalia, echopraxia, and other peculiarities of voluntary movement. Tics and other sudden, non-rhythmic movements, often with self-injury, occur commonly in catatonic patients and may qualify as additional catatonic symptoms [3,4].

During the last 15 years, catatonia has been further delineated in children, adolescents, and young adults across a wide range of disorders, including autism [5-7]. Catatonia has been increasingly recognized as a comorbid syndrome of autism at a rate of 12-17% in adolescents and young adults with autism spectrum disorders [8,9] and with other intellectual disabilities [10,11].

An overview of pediatric and adult disorders, in which catatonia can emerge, is given in the Table 1, showing that catatonia occurs in patients with psychotic, affective, drug-induced, medical, autistic, developmental, tic, and psychogenic disorders. In some catatonic patients, no other medical or psychiatric diagnosis can be clearly made [3,12].

There are several accounts of children and adolescents in whom catatonia developed after severe traumatic events, both in past and recent literature [40]. German psychiatrist Karl Kahlbaum [41], who coined the term catatonia in 1874, gave trauma a central role in catatonia in many young adult cases. Kanner [35] described children with psychogenic catalepsy. Anaclitic depression [36,37], a condition found by Spitz in deprived institutionalized children, meets criteria for stuporous catatonia. Leonhard [42] considered lack of communication

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<th>Developmental Disorders</th>
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<td>Autism Disorder [13-16]</td>
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<td>Childhood Disintegrative Disorder [12,17]</td>
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<td>Mental Retardation [5] including Down Syndrome [18]</td>
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<td>Prader Willi Syndrome [10]</td>
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<th>Medical and Neurological Disorders</th>
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<td>Catatonia due to a general medical condition (brain structural damage, seizures, metabolic, endocrine, and autoimmune disorders) [19-23]</td>
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<th>Psychiatric Disorders</th>
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<td>Psychotic Disorders [24]</td>
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<td>Mood Disorders [24]</td>
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<td>Substance-induced disorders [19]</td>
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<td>Medication-induced movement disorder (NMS) [25,26]</td>
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<td>Tourette’s Syndrome [3,6,27]</td>
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<th>Miscellaneous Conditions</th>
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<td>Anti-NMDA (N-methyl-d-aspartic acid) receptor encephalitis [28-32], PANDAS (Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal Infections) [29,32], encephalitis lethargica [33], and other aseptic encephalitides [34]</td>
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<td>Kleine Levin Syndrome [12]</td>
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<td>Psychogenic catalepsy [35]</td>
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<td>Anaclitic depression [36,37]</td>
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<td>Pervasive Refusal Syndrome [38,39]</td>
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Table 1: Pediatric and adult disorders in which catatonia can emerge.
with the mother or substitute mother as an important risk factor for childhood catatonia. Children who experience emotional and physical trauma sometimes develop catatonia [43]. The descriptions of contemporary traumatized refugee children with a syndrome labeled Pervasive Refusal Syndrome are those of children with classic catatonic syndromes [38,39].

Materials and Methods

Review of literature and case-reports on the role of fear in the development of catatonia in autism, and its treatment.

Results

Catatonia in autism and its treatment

Catatonic symptoms such as mutism, stereotypic speech, echolalia, stereotypic or repetitive behaviors, posturing, grimacing, rigidity, mannerisms and purposeless agitation feature prominently in autism. Some patients experience a sharp increase of these symptoms, often in adolescence, and qualify for a diagnosis of catatonia [9,44].

Many such youngsters suffer extreme physical compromise, including inability to move, eat or void, development of dangerous cardiovascular and thermoregulatory instability as well as onset of repetitive, tic-like, self-injury with risk of further tissue and organ damage [14,16,45-57].

Electroconvulsive therapy (ECT) resolved catatonia in several of these patients with improvement in interpersonal behavior and ability to attend school and social events [16,46,49,53,56-57]. Other, usually milder, cases have benefitted from the use of lorazepam [55], and social, behavioral, and psychological interventions [54].

A growing literature indicates that ECT is safe and efficacious in the acute treatment of catatonia in autism. Maintenance ECT lasting months or years may be pursued to maintain remission [58], and early data supports stability of longitudinal neuropsychological testing in such patients [59].

Case-Vignettes

Shah and Wing [54] found that ongoing stressful experiences often precede the development of catatonia in autistic people. Life events, medical events, onset of puberty, the loss of routine and structure, experiences of loss, conflicts with parents, caregivers, or peers, and discrepancies between the higher functioning autistic individual's capabilities and the expectations of parents, seem to be able to precipitate in catatonia as illustrated in their case-vignette (Case one).

Case one [54]

A 23-year-old man previously diagnosed with autism presented with progressive catatonia over the last 4 years since leaving a special school for autistic children and becoming increasingly socially isolated while staying at home. He used to be fully independent in all aspects of self-care, but had become totally dependent on his mother. The morning routine of getting up, washing and dressing was taking up to five hours. He frequently became ‘frozen’ and each small meal was lasting for hours. He was severely underweight and still losing more weight. He hardly talked at all. When asked questions, he took a long time to respond and even then was able to answer only in monosyllables. He whispered the answers very softly. He seemed unable to lift his head to make eye contact or to interact. He had a fixed expression on his face and was not able to acknowledge people in any way. He was spending most of his waking time on a sofa in the living room, in a fixed stooped posture with his head bowed and his arms hanging by his sides. He tried to respond to instructions but each movement seemed agonizingly difficult. Before each action, his body twitched and jerked, he blinked continuously and made repetitive movements with his mouth. Interventions consisted of involvement of the local services for people with learning difficulty, support and psycho-education to the family, and implementation of an intervention plan based on principles described by Shah and Wing [54], slowly improving his condition. As he gradually regained his mobility, speech and independence, he was able to be placed in a structured day activity centre.

Vignettes of two other published cases [60,61] are presented which abusive and traumatic events precipitated in catatonia in autistic youngsters.

Case two [61]

A 16-year-old adolescent male with a diagnosis of Asperger Syndrome and long history of being abused by his mother, developed catatonia characterized by mutism, immobility, catalepsy, and mannerisms after the illness of his grandfather to whom he was close. The patient responded dramatically to diazepam. The case illustrates the importance of assessing catatonia in patients with autism spectrum disorders who present with motor disturbances in the presence of abusive or traumatic events, and possible diagnostic confusion with catatonic schizophrenia or Reactive Attachment Disorder. The patient's catatonic reaction struck the authors as “death mimicry due to the subjective perception of a life-threatening situation” for which treatment with benzodiazepines and ECT was indicated.

Case three [60]

An 11-year-old boy with high-functioning autism, previously well-adjusted and verbal, developed catatonia characterized by psychomotor retardation, muteness, food refusal, incontinence, and withdrawal, following the sudden death of his father. The boy was treated with antidepressant and antipsychotic medications without benefit, but responded, with full resolution of catatonia, to a test dose of 1 mg of lorazepam after which he was able to return to school. He stayed well on 1 mg of lorazepam daily.

Discussion

There are no empirical studies in the literature that address the role of fear in the development of catatonia in autism. However, there are several accounts of children and adolescents in whom catatonia developed after severe traumatic events [40]. Case-reports support that ongoing stressful experiences often precede the development of catatonia in autistic people. Future studies should assess fear due to psychological or medical trauma as risk factor for catatonia. Underreporting of psychological trauma and under recognition of the diagnosis of catatonia are challenging clinical and research issues.

There is a support from case-reports that catatonia in autism is a treatable syndrome, often requiring the use of benzodiazepines, ECT, and maintenance ECT for safe reversal. Social, behavioral, and psychological interventions for catatonia aim to reduce anxiety. Therefore, further treatment studies are warranted.

Next, factors that could increase the risk for catatonia due to fear in autistic people are examined, and biological underpinnings of catatonia as motor reaction to fear are explored.

Fear by everyday life in autism

Robin Allott [62] graphically described the fear provoked by everyday life due to autistic impairment. "Language, vision, and motor
control all go together; language is the supreme medium of empathy and language almost certainly plays the major role in making possible consciousness, self-awareness; language allows one to construct a model of one’s world, to create rational expectancies, particularly about the behavior of others. For someone with no reliable pattern of expectation about the ‘world’, every moment of life becomes like wandering through a Chamber of Horrors, unknown and unexpected horrors.

Autistic people may be particularly vulnerable to intense fear [63,64] due to deficits in communication and social skills, low cognitive abilities, and heightened sensory sensitivity. In a study of autistic adults [63], levels of anxiety, as assessed by caregivers, were threefold higher than in a comparison group of individuals with intellectual disabilities. In the autistic group, levels of anxiety correlated with levels of stress.

There is a growing evidence that autistic children have deregulated hypothalamic-pituitary-adrenal stress responses, compared with control groups [65] and abnormal central GABA function [66] interfering with the central integration of hypothalamic-pituitary-adrenal stress responses in the basal forebrain and hypothalamus [67].

Observations that catatonia follows overwhelming anxiety due to trauma or perceived danger, the positive response of catatonia to anxiolytics such as benzodiazepines or barbiturates, and psychogenic theories of catatonia [68] are particularly applicable to people with autism due to their increased social, cognitive, and sensory vulnerabilities.

**Catatonia: The ultimate motor response to fear**

Catatonia has been called “the ultimate response to fear” [69], representing a common final pathway in the response to impending doom, analogous to the animal defense strategy of tonic immobility or freezing [70].

Porges [71-73] has proposed the Polyvagal Theory in which the human freeze or immobility response, with ensuing behavioral and metabolic shutdown, represents the most primitive response to perceived imminent danger, in a phylogenetically organized hierarchy of autonomic functions, when fight/flight reactions fail or are not available.

A separate set of unmyelinated fibers of the vagus nerve projecting to the dorsal motor nucleus of the vagus is thought to mediate this immobilization. The freeze response is adaptive in reptiles but potentially lethal in humans due to prolonged behavioral (death feigning and passive avoidance) and metabolic shutdown (negative nitrogen balance, vasovagal responses).

The Polyvagal Theory has been tested in autistic people in regards to impairment of pro-social neuronal circuits consisting of myelinated vagal fibers projecting to the nucleus ambiguous [74], but not in regards to increased risk for immobilization or catatonia. There are no studies on the role of the vagus nerve in catatonia.

The lack of self-reported anxiety in many catatonic patients, including those with autism [54,58], does not rule out catatonia as a motor reaction to anxiety, given the separate circuitry for emotional and motor responses to perceived danger. Self-reports of anxiety may be further compromised in autistic patients with their known difficulties in expressing inner states in words. Amnesia for the duration of catatonia is another puzzling but common phenomenon that may be due to extreme levels of stress and arousal, similar as in traumatic dissociative amnesia [75].

Further human study of the immobilization response [70,76] and Polyvagal Theory [73] promises to increase our understanding of the role of trauma in catatonia, to clarify the evolution-based effects of perceived danger on the relation between visceral states and social engagement and disengagement, and to elucidate the mechanism whereby extreme fear by perceived danger can induce such profound and perilous reaction like catatonia often requiring the use of benzodiazepines or ECT for safe reversal [77].

**Conclusions**

The current article shed light on the following:

1. Catatonia may represent an extreme fear reaction to perceived danger.
2. Autistic people may be particularly vulnerable to catatonia due to social, cognitive, and sensory deficits.
3. People with autism who develop catatonia should be assessed for stressful, traumatic, or abusive events in family and social environments, in addition to medical causes of catatonia.
4. Catatonia is a treatable syndrome, often requiring the use of benzodiazepines, ECT, and maintenance ECT for safe reversal, when occurring in people with autism.
5. Social, behavioral, and psychological interventions for catatonia aim to reduce anxiety.
6. Further studies are warranted in autistic people, measuring states of anxiety in response to various stressors, and assessing their relationship to catatonia, and applying various treatments including benzodiazepines, electroconvulsive therapy, and psychosocial interventions, in those with catatonia.

**References**


