ABSTRACT:

Catatonia is a pathological entity with deep historical roots and it has received many interpretations over the time. The association between catatonia and autism still raises many questions, which makes many authors attempt to find answers based on clinical trials.

Catatonia is a syndrome of specific motor abnormalities closely associated with disorders in mood, affect, thought, and cognition. The principal signs of the disorder are mutism, immobility, negativism, posturing, stereotypy, and echo phenomena.

Adolescents and young adults with autism could present during lifetime a psychiatric episodic disorder, including catatonia. There are studies who have tried to analyze the importance of predicting factors like IQ, age, gender, history of seizure disorder and also to find a correlation between catatonic episodes and development of autistic persons.

Key words: catatonia, autism, child & adolescent

REZUMAT:

Catatonia reprezintă o entitate psihopatologică cu adânci rădăcinii istorice și care a primit multiple interpretați de-a lungul timpului. Asocierea dintre catatonia și autism ridică încă multe semne de întrebare, ceea ce face ca mulți autori să încerce să găsească răspunsurile pe baza studiilor clinice.

Catatonia este un sindrom caracterizat de anomalii motorii specifice, strâns asociate cu tulburări ale dispoziției, afectivității, gândirii și cunoştinţei. Semnele principale ale tulburare sunt mutism, imobilitate, negativism, catatonia rigidă, stereotipii și ecofenomene.

Adolescenții și adulții tineri cu autism ar putea prezenta în timpul vieții o tulburare psihiatrică episodică, inclusiv catatonia. Există studii care au încercat să analizeze importanța factorilor predictori, cum ar fi IQ-ul, vârsta, sexul, antecedentele de tulburări convulsive și, de asemenea, să găsească o corelație între episodul catatonic și dezvoltarea ulterioră persoanelor cu autism.

Cuvinte cheie: catatonia, autism, copil și adolescent.

Catatonic stupor is one of the most dramatic psychiatric presentations, but is becoming increasingly rare in our days. Catatonia is a syndrome of specific motor abnormalities closely associated with disorders in mood, affect, thought, and cognition. The principal signs of the disorder are mutism, immobility, negativism, posturing, stereotypy, and echo phenomena.

Since 19th century, the manifestations of catatonia have been given individual names as different authors identified features that they considered unique.

The concept of catatonia was first formulated and the illness named by Kahlbaum in 1874. In a 104 page book entitled “Die Katatone oder das Spannungsräusche”, Kahlbaum described his experience with 26 patients in both the stuporous and excited forms of the illness. He defined 17 signs of the syndrome following the diagnostic rules set down by Sydenham, more than a hundred years earlier. Kahlbaum’s catatonia had previously been recognized as stupor, and the French called the lack of communication ‘stupidite’. (Fink et al., 2010) The symptoms were not unfamiliar and were pointed by subsequent generations of clinicians across a wide range of different disorders.

Rigid posture, mutism, negativism, and catalepsy initiated an illness that was soon followed by the hyperkinetic phenomena of stereotypes, verbiage, and excitement. Catatonia was “a temporary stage or a part of a complex picture of various disease forms.” Kahlbaum also compared catatonia to dementia paralytic, a disorder that dominated the psychiatric practice at the time. (Fink et Taylor, 2003)

In his 1913 textbook, Karl Jaspers, portrayed catatonia as an illness sui generis characterized by opposing pairs of symptoms (e.g. negativism vs. automatic obedience). Jaspers’ main interest was in the psychology of catatonia, which he found unknowable: ‘Sometimes it seems as though the patient is like a dead camera: He sees everything, hears everything, understands everything and yet is capable of no reaction, of no affective display, and of no action. Even though fully conscious he is mentally paralyzed.” (Fink et al., 2010)

The most profound commentary on Kahlbaum’s work

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was that of Emil Kraepelin. He folded Kahlbaum’s descriptions of catatonia and hebephrenia into his concept of dementia praecox. Kraepelin’s voice has dominated psychopathological thought to the present day and is the basis for the classification of catatonia in DSM and ICD systems. (Dhossche, 2006, Fink et al., 2010) Adolf Meyer, Smith Eli Jellife, and William Alanson White, who led American psychiatry, viewed schizophrenia and especially its catatonic form as evidence of the psychological basis for the psychoses. Such views became the basis for the 1952 DSM classification that described abnormal behaviors as reactions to psychological and physical stressors, and not as defined syndromes. He interpreted catatonic signs as mental blocking without a structural basis. (Fink et Taylor, 2003).

Bleuler, using the same concepts, described mutism, negativism, and rigidity as “generalized and persistent blocking – an exaggeration of the phenomenon seen in healthy individuals when they are overwhelmed by emotional disturbance.” (Dhossche, 2006)

Stuporous catatonia is characterized by varying combinations of mutism, immobility and waxy flexibility; associated features include posturing, negativism, and automatic obedience and “echo” phenomena. Mutism ranges from complete to partial: some patients may mumble or perhaps utter brief, often incomprehensible, phrases. Immobility, likewise, ranges in severity: some patients may lie in bed for long periods, neither moving, blinking or perhaps even swallowing; others may make brief movements, perhaps to pull at a piece of clothing or to assume a different posture. Waxy flexibility, also known by its latin name, *cerea flexibilitas*, is characterized by a more or less severe “lead pipe” rigidity combined with a remarkable tendency for the limbs to stay in whatever position they are placed, regardless of whether the patient is asked to maintain that position or not. Posturing is said to occur when patients spontaneously assume more or less bizarre postures, which are then maintained. Negativism entails a mulish, intractable and automatic resistance to whatever is expected, and may be either “passive” or “active”. Passively negativistic patients simply fail to do what is asked or expected of them, while active negativism manifests in doing the opposite of what is expected. Automatic obedience, as may be suspected, represents the opposite of negativism, with affected patients doing exactly what they are told, even if this should place them in danger. Echo phenomena represent a kind of automatic obedience: in echolalia patients simply repeat what they hear and in echopraxia they mimic the gestures and activity of the examiner. (Dhossche et al., 2006)

Catatonia is seen in its most severe variant in individuals who ‘freeze’ as they move through a room, and are halted in mid-action. They literally remain in a particular posture for minutes, sometimes even hours. (Gillberg, 2002)

Catatonia is a syndrome of altered motor behavior accompanying many general medical and neurologic disorders. It is more frequently found among patients diagnosed with mania, depression, and neurotoxic syndromes than among those with schizophrenia. (Fink et al., 2010)

The range of primary diagnoses which catatonia is associated with is similar to that described in adults, but in addition, catatonia is reported in patients with the defined childhood disorders of autism, genetic syndromes, and mental retardation.

Dhossche and Bouman summarized the diagnoses in 30 adolescents and children with catatonia described in the literature between 1966 and 1996. Eleven patients had been diagnosed as suffering from atypical or brief psychotic disorders, 10 had a neurologic or a general medical condition (epilepsy, drug intoxication, viral infection), six had a mood disorder, and three had schizophrenia. Of the patients with atypical psychoses, three were seen also to have a developmental disorder (two with infantile autism; one with mental retardation). Stupor or catalepsy (27/30), mutism (26/30), posturing/ grimacing/stereotypy (16/30), echolalia or echopraxia (4/30), and excessive motor activity (4/30) were the catatonic features that met DSM-IV specified criteria. (Dhossche and Bouman, quoted by Fink et Taylor, 2003)

Other syndromes with signs of catatonia have been described in young patients. British authors delineate a pervasive refusal syndrome, when the patients are mute, withdrawn, and apathetic, with a paucity of interactions. The syndrome is assumed to result from psychological trauma, and the treatment offered is individual and family psychotherapy. (Lask et al., Quoted by Fink et Taylor, 2003).

A syndrome of idiopathic recurrent stupor (IRS) is described in patients who exhibit periodic stupor without toxic, metabolic, or structural brain damage. Some patients have exhibited fast 14 Hz background activity in the EEG. The relationship of IRS to the syndromes of catatonia, autism, pervasive refusal syndrome, and varieties of mental retardation is a puzzle (Mac Keith, quoted by Fink et Taylor, 2003), that interests child and adolescent psychiatrists.

Catatonia is a motor phenomenon that sometimes becomes a major problem in children with Autism Spectrum Disorder (ASD). The most common time period for its onset is in preadolescence and adolescence, and it is not uncommon (Gillberg, 2000), although the introduction of antipsychotics has reduced its incidence.

In 1998 Lorna Wing noted that catatonia may occur...
in adolescents and young adults with autistic disorders, of any ability level. Few years later, Wing and Shah (2000) operationally defined catatonia in individuals with ASDs. In their definition, four features were taken up—that is, (1) increased slowness affecting movements and verbal response, (2) difficulty in initiating and completing action, (3) increased reliance on physical or verbal prompting by others, and (4) increased passivity and apparent lack of motivation. As often-associated symptoms, they referred to (5) reversal of day and night, (6) parkinsonian features (tremor, eye-rolling, dystonia, odd stiff posture, freezing in postures, etc.), (7) excitement and agitation, and (8) an increase in repetitive and ritualistic behavior.

Wing and Shah have suggested that about 10–20% of all individuals with autism and IQs above 50 may have moderate to marked features of catatonia. They reported that the age-of-onset of catatonia ranged from 10 to 30 years of age, with a peak at 15–19 years. However, it has been suggested that catatonia is under-recognized and under-diagnosed. The risk of developing catatonia appears to be greatest among those who have an early history of language delay and who belong in the passive subgroup of autism spectrum disorders. Parents of the individuals in Wing and Shah’s study, who exhibited catatonic-like deterioration, reported a variety of precipitating events, ranging from school examinations to bereavement. The authors also noted that there were no significant differences in the age, gender, IQ, history of seizure disorder, or in the diagnostic subgroup for autism noted between the individuals showing symptoms and the comparison control group.

A study from 2005 investigated the prevalence of episodic psychiatric disorders (including delusions, hallucinations, catatonia, etc) in teenagers with learning disabilities, with and without autism. It was found that teenagers with learning disabilities and autism have higher rates of episodic psychiatric disorders than those with learning disabilities alone. Significantly more individuals with autism had a lifetime episodic disorder. They also concluded that the deterioration may not be specifically associated with autism, but rather with learning disabilities in general and the addition of predisposing factors, such as family history and/or comorbid psychiatric disorder. (Bradley et Bolton, 2006)

Special attention needs to be paid to persons with pervasive developmental disorders (PDD). As they often have language impairment and may be mute, the presence of mutism should not be deemed a sign of catatonia unless it is episodic. Other features, especially posturing, echolalia/echopraxia, and stereotypy need to be present. Catatonia should also be considered when adolescents have episodes of irritability and excitement followed by periods of mutism, catalepsy, and negativism. (Fink et Taylor, 2003)

Most cases of catatonia are precipitated by psychiatric causes such as depression and psychosis. People with ASDs may experience a disproportionate number of negative life events such as bullying and social exclusion at school, particularly during the adolescent years. Although mood disorders can be diagnosed at any level of intellectual ability, cognitive ability and the autism phenotype itself will both have a modifying effect on presentation. (Ghaziuddin et al., 1992) A depressive disorder may present as an increase in social withdrawal or stereotypic movements, or manifest as catatonia. (Hollander et Anagnostou, 2007)

Occasionally, catatonia may occur repeatedly in the same patient, a condition known as periodic catatonia (Wing & Shah, 2000). However, there were also a few cases in which catatonia repeatedly aggravated over short spans of time. Catatonia in ASDs may be considered an epiphenomenon of ASDs or a manifestation of comorbidity in adolescence or early adulthood. (Dhossche et al., 2006)

Dhossche and his collaborators have compared the results of three studies and have confirmed the comparatively high rates of catatonia-like features and catatonia-like deterioration in people with ASDs. Apart from the tendency to passivity in social interaction and the impaired expressive language, no features that predicted the likelihood of catatonia-like deterioration were identified. The studies have not thrown any light on the relationship between these two clinical pictures apart from the fact that they both comprise unusual patterns of movement, speech, and behavior. It could be suggested that the presence of so many catatonia-like features makes people with ASDs especially vulnerable to catatonia-like deterioration. However, clinical experience has shown that such deterioration can occur in adolescents or adults, who are very high functioning and who, before the deterioration, had few if any catatonia-like features even in childhood.

Furthermore, the majority of people with ASDs do not develop catatonia-like deterioration even if they have had many catatonia-like features. (Dhossche et al., 2006) Because of the way the data were collected, it’s impossible to examine in detail the relationship between the history of catatonia-like features and the development of catatonia-like deterioration. Billstedt and his collaborators have published in 2005; a population based follow-up study of 120 individuals aged 17–40 years with ASDs diagnosed in childhood. They found that 13 (12%) had severe motor initiation problems similar to Wing’s criteria for catatonia.
like deterioration. (Dhossche et al., 2006) The association of autistic disorders with catatonia has also caused confusion with schizophrenia but clinical impression suggests that catatonia-like deterioration is related to events that are experienced as stressful. (Hollander et Anagnostou, 2007)

The psychological and biological correlation of catatonic regression in people with and without autism are unknown (Fink and Taylor, 2003; Wing and Shah, 2000). Acute deterioration should prompt investigations for underlying medical and neurological disorders, particularly seizure disorders. Fink and Taylor (2003) find meager evidence for a direct connection between EEG measures and catatonic symptoms. Diffuse slowing has been reported in patients in catatonic stupor. Others have reported a dysrhythmic EEG in catatonia, consistent with nonconvulsive status epileptics that resolved when the catatonia resolved. (Dhossche et al., 2006)

In a small number of cases, catatonia may be associated with medical factors, such as chronic infections (Gilberg, 2002). In addition, there is growing evidence that the use of antipsychotic medications may increase the risk of catatonia and of neuroleptic malignant syndrome, a related condition. (Wing and Shah, 2000) In some cases, the cause of catatonia may not be apparent.

Etiological explanations for catatonia include a genetic predisposition, especially for periodic catatonia. Genetic linkage studies have found a shared susceptibility region, 15q15-q21, for both autism and catatonia. Research appeared to support a single gene model and an autosomal dominant mode of inheritance with reduced penetrance (Stober, 2004 – quoted by Dhossche et al., 2006)

Similarities between autism and catatonia include also abnormal GABA function and small cerebellar structures. Numerous investigators have posited a key role for central dopaminergic hypoactivity in triggering catatonia. The onset of catatonia coincides with a reduction in dopaminergic activity within the basal ganglia–thalamocortical circuits. (Rogers, 1991; Fricchione et al., 2000; Dhossche et al., 2006) Five circuits connecting the basal ganglia with their associated areas in the cortex and thalamus have been identified and are named according to their function or cortical site of origin. They include the “motor circuit,” the “oculomotor circuit,” the “dorsolateral prefrontal circuit,” the “lateral orbitofrontal circuit,” and the “anterior cingulate–medial orbitofrontal circuit.” The anterior cingulate–medial orbitofrontal subcortical circuit may mediate diminished arousal, mutism, and akinesia in simple catatonia. (Taylor, 1990; Mann et al., 2003)

In general, the prognosis for the acute catatonic phase seems to be good, but the long-term prognosis probably depends on the underlying cause of the catatonia. (Sundararajan, 2007) If a patient with catatonia does not eat or drink for prolonged periods, this will lead to dehydration and its attendant complications. The immobility of catatonia may increase the risk of deep-vein thrombosis (Morioka et al, 1997). McCall and his collaborators have highlighted the increased risk of death due to pulmonary embolism in patients with persistent catatonia; such deaths occurred only after the second week of catatonia, often without warning. (McCall et al., 1995) During the phase of catatonic excitement, the patient may pose a significant risk of harm to self and others. (Sundararajan, 2007)

Autism and catatonia are important psychopathological dimensions with deep historical roots. Autism spectrum disorder and catatonia are conditions that are on the borderline and overlap with many clinical specialties: psychiatrists, neurologists, pediatricians, psychologists, and other child specialists.

Catatonia should be assessed in any patient with ASDs when there is an obvious and marked deterioration in movement, pattern of activities, self-care, and practical skills, compared with previous levels, through a comprehensive diagnostic evaluation of medical and psychiatric symptoms.

REFERENCES: