What exactly is catatonia in children and adolescents

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Summary

The current study is a review of the literature on catatonia syndrome with focus on children and adolescent’s specificity. Previous catatonia conceptualizations were significantly modified in the newest classification systems. Catatonia may be considered either a separate syndrome or a specifier of the course of other psychiatric disorders. Although diagnostic criteria for children and adolescent do not differ from those for adults, the clinical presentation and course may not be the same. In this age group relatively common are somatic conditions taking the form of catatonia. There is a growing body of literature focused on catatonia in the course of pervasive developmental disorder. On the other hand, pervasive refusal syndrome and lethal catatonia are discussed in the literature, but they are not present in the classification systems. In the current paper basic treatment guidelines were also described. First-line treatment is the use of benzodiazepines and electroconvulsive therapy. The diagnosis and treatment of catatonia is of great practical importance. While improper diagnosis and non-optimal treatment may have fatal consequences, in the case of proper diagnosis an effective treatment may be administered.

Key words: catatonia, classification of mental disorders, autism spectrum disorder, autoimmune encephalitis

Definition

Derived from the Greek language, the word ’katatonia’ means completely (katá-) taut (-tonikós). Catatonia is a syndrome in which movement disturbances are dominant. An increased (hyperkinetic form) or reduced (hypokinetic form) motor activity may be observed. This state is accompanied by qualitative disturbances of physical activity and significantly reduced or no contact with the patient [1].
Development of the concept

It is widely acknowledged that catatonia was for the first time defined and described by Karl Ludwig Kahlbaum – first in a lecture given in 1868, then in print, in 1874 in a small monograph Die Katatonie oder das Spannungsirresein (Catatonia or crazy tension) in Psychische Krenkheiten (Mental Illness) [1]. It is worth mentioning, however, that the first description of the condition later known as lethal catatonia is dated to 1832 [as cited in: 2].

Kahlbaum described catatonia as a set of symptoms that can occur in the course of different conditions – mainly in mood disorders. The inclusion of catatonia (1904, Emil Kraepelin) to the clinical picture of dementia praecox results in the understanding of catatonia mainly as a schizophrenic syndrome. Also Eugene and Manfred Bleuler, working in the first half of the 20th century, distinguished it as a subtype of schizophrenia [1].

Later, Leonhard and his contemporary researchers in the process of differentiation numerous subtypes of psychotic disorders distinguished a motility psychosis among cycloidal psychoses, as well as systemic and unsystematic catatonic schizophrenia among schizophrenia. According to Neumärker [3], Leonhard considered catatonic syndrome to be a separate group of disorders. Although in his initial work Leonhard considered catatonia in children and adolescents to be associated with schizophrenia, in later years he emphasized the need of careful differentiation between catatonia and – as we would say today – neurodevelopmental disorders.

The research in the 1970s and 1980s showed that catatonic syndromes may occur in the course of affective disorders (from 30% to about half of catatonia cases – much more frequent than in the course of schizophrenia: 10–20% of cases) and in somatic diseases (about 20%) [4, 5]. This observation was discordant with classification systems until publishing the DSM-5 classification in 2013 [6].

The ICD-10 classification is still used in Poland. Catatonia may be diagnosed in catatonic schizophrenia or in organic catatonic disorder. There are, however, three more ICD-10 chapters where characteristic mobility symptoms – which are the essence of the matter in catatonia – are discussed. In the chapter concerning mood disorders it may occur in F31.5, F32.3, F33.3 – severe depressive episode with psychotic symptoms). In ICD-10, depressive stupor is considered to be a form of psychotic depression. In the next chapter, the code F44.2 – dissociative stupor – is described. Nevertheless, dissociative stupor following an experienced trauma is coded as F43.02 – acute stress reaction, severe. Moreover, in the chapter Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified the code R40.1 refers to stupor [7].

In the DSM-5 classification, catatonia may be diagnosed either as a separate syndrome or as a classifier in the course of other mental conditions like schizophrenia, mood disorders but also in the course of conditions not linked with catatonia in the past [6].

In ICD-11, catatonia is described as a separate parent category, on equal footing with schizophrenia. It includes 3 subcategories, all of each use the same definition
of catatonia (Table 2). In the eleventh version of ICD, catatonic schizophrenia no longer exists. Catatonia was also included into a category Symptoms, signs or clinical findings, not elsewhere classified, including concepts and their definitions that can be used to determine important aspects of the patient’s clinical picture, regardless of what specific mental disorders have been diagnosed. Therefore, catatonia may be diagnosed irrespectively of the primary diagnosis, as a clinical presentation specifier [8–10].

It is worth mentioning that there are also diagnostic codes Catatonia induced by substances or medications in the Catatonia chapter and secondary catatonia coded elsewhere. Catatonic symptoms present during or shortly after the consumptions of legal or illegal drugs should be classified under subcategory Catatonia induced by substances or medications. Symptoms which fulfil the criteria of catatonia, without connection to delirium or other mental or behavioural disorders, which could be explained by patient’s medical history, physical examination or laboratory findings fall into parent category: Secondary mental or behavioural syndromes associated with disorders or diseases classified elsewhere.

Clinical presentation

The currently defined catatonic syndrome is described as a set of symptoms related mainly to the motor area [11, 12]. Depending on the source, we can find descriptions from five to at least ten main symptoms of catatonia, among which the most frequently described are:

• mutism – complete lack of verbal communication, not necessarily connected with immobility;
• posturing – temporary stopping of physical activity;
• immobility – long-term retention of movement activity, it is possible to react to external stimuli;
• stupor – immobility connected with mutism, no response to external stimuli;
• negativism – the patient resists the examination, using adequate strength – e.g., during a neurological examination he/she does not allow to change his/her position, but he/she does not change it himself/herself;
• agitation – pointless motor activity unprovoked by any external factor.

Other symptoms of catatonia, considered almost pathognomonic, include:

• catalepsy – specific stiffening of the muscles, combined with the stiffening of the body and the position of the limbs and neck bending, also in unnatural positions, related to the adoption of bizarre positions; waxy flexibility may be an element of catalepsy;
• waxy flexibility – the patient maintains the position given by the examiner;
• automatic obedience – the patient unconsciously, without reflection and automatically performs the instructions of the investigator, also those that should seem
strange or potentially unpleasant, e.g., patient responds to the command: “Please stick out the tongue, I will pin the pin to it”

• passive obedience – the patient, against the verbal command, submits to a delicate movement made by the investigator who changes his/her position, the patient gives in and maintains a new position for some time (e.g., the investigator lifts the patient’s arm, which he/she previously recommended to keep on his/her knee);

• stereotypies – repetitive seemingly deliberate but non-functional movements or linguistic stereotypies;

• ambitendency – a frequently described example of catatonic ambitendency is when the patient reaches out a hand with a greeting, with the commentary “do not give me a hand, I do not want you to give me a hand”;

• echo phenomena – echolalia (unnecessary repetition of heard words) and echopraxia – repetition of observed movements;

• mannerisms – purposeful but bizarre movements like saluting random people, simulating holding a gun;

• disorganization of behaviour – loss of the possibility of performing targeted actions, which makes basic self-care impossible;

Most of these symptoms have been included in the diagnostic criteria presented in Tables 1 and 2.

Catatonia is a syndrome that can have a diverse clinical picture. The most characteristic symptoms of catatonia like waxy flexibility and bizarre poses may not be present. Moreover, catatonia may be associated with arousal, immobility or stupor. That all sometimes makes catatonia diagnosis difficult [13].

Table 1. **Criteria for catatonia according to ICD-10**

<table>
<thead>
<tr>
<th>F20.2 Catatonic schizophrenia</th>
</tr>
</thead>
<tbody>
<tr>
<td>A. The general criteria for a diagnosis of schizophrenia (F 20.0–F20.3) must be satisfied, although in the first stage, if patient is uncommunicative, adequate evidence of the presence of symptoms may not be obtained</td>
</tr>
<tr>
<td>B. One or more of the following behaviors should dominate the clinical picture for at least 2 weeks:</td>
</tr>
<tr>
<td>1. stupor or mutism;</td>
</tr>
<tr>
<td>2. excitement;</td>
</tr>
<tr>
<td>3. posturing;</td>
</tr>
<tr>
<td>4. negativism;</td>
</tr>
<tr>
<td>5. rigidity;</td>
</tr>
<tr>
<td>6. waxy flexibility;</td>
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<tr>
<td>7. command automatism</td>
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</table>

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<tr>
<th>F06.1 Organic catatonic disorder</th>
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<tbody>
<tr>
<td>The general criteria for a diagnosis of other mental disorders due to brain damage and dysfunction and to physical disease</td>
</tr>
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One of the following:
1. Stupor
2. Negativism
3. both (shifting rapidly and unpredictably from hypo – to hyperactivity)

Comment: Other catatonic phenomena that increase confidence in the diagnosis are: stereotypies, waxy flexibility and impulsive acts. The delirium should be delimitated, although it has not been conclusively determined whether an organic catatonic state may occur in clear consciousness or whether it is always a manifestation of delirium.

Table 2. ICD-11 and DSM-5 criteria for catatonia

<table>
<thead>
<tr>
<th>Definition of catatonia</th>
<th>DSM 5</th>
<th>ICD 11</th>
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<tbody>
<tr>
<td>The presence of three or more of the following:</td>
<td></td>
<td>The presence of several of the following:</td>
</tr>
<tr>
<td>1. Stupor</td>
<td></td>
<td>1. Extreme slowing or absence of motor activity</td>
</tr>
<tr>
<td>2. Catatlepsy</td>
<td></td>
<td>2. Mutism</td>
</tr>
<tr>
<td>3. Waxy flexibility</td>
<td></td>
<td>3. Purposeless motor activity unrelated to external stimuli</td>
</tr>
<tr>
<td>4. Mutism</td>
<td></td>
<td>4. Assumption and maintenance of rigid, unusual or bizarre postures</td>
</tr>
<tr>
<td>5. Negativism</td>
<td></td>
<td>5. Resistance to instructions or attempts to be moved</td>
</tr>
<tr>
<td>6. Posturing</td>
<td></td>
<td>6. Automatic compliance with instructions</td>
</tr>
<tr>
<td>7. Mannerisms</td>
<td></td>
<td></td>
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<tr>
<td>8. Stereotypies</td>
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<td></td>
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<tr>
<td>9. Agitation</td>
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<td></td>
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<tr>
<td>10. Grimacing</td>
<td></td>
<td></td>
</tr>
<tr>
<td>11. Echolalia</td>
<td></td>
<td></td>
</tr>
<tr>
<td>12. Echopraxia</td>
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</tbody>
</table>

Duration | Not specified | Not specified |

Classification | | Parent category: Catatonia |
| | Subcategories: | |
| 1. Specifier “with catatonia” for: | 1. 6A40 – Catatonia associated with another mental disorder (schizophrenia, mood disorders, autism spectrum disorder, others, i.e., Prader–Willi syndrome, tic disorder) |
| | – Psychotic disorder (schizophrenia, schizoaffective disorder, schizopreniform disorder, brief psychotic disorder) | |
| | – neurodevelopmental disorder | |
| | – mood disorders – major depressive episode and bipolar disorder | |
| | – other mental disorders | |
| 2. Catatonia due to a general medical condition | 2. 6A41 – Catatonia induced by psychoactive substances, including medications |
| 3. Catatonia disorder not otherwise specified | 3. 6A4Z – Catatonia, unspecified |

Parent category: Secondary mental or behavioral syndromes associated with disorders or diseases classified elsewhere |

Subcategory: |

1. 6E69 – Secondary catatonia syndrome
Etiology and pathomechanism of catatonia syndrome

Catatonia remains a condition of unknown etiology. Multiple disorders may be associated with catatonia syndrome, which may suggest complex nature of this issue. Organic catatonia, which is discussed later in the paper, is relatively common in children and adolescents. Role of genetic factors in catatonia was also suggested [4].

Pathomechanism of symptoms’ clinical presentation has also been studied. In catatonia both motor and volitional functioning is affected. One of main proposed hypotheses is hyperactivity of glutaminergic system and hypoactivity of GABA-ergic system in the central nervous system. Clinical efficacy of drugs targeting GABA-A receptor and antagonists of the glutamatergic receptor confirms the important role of the above systems in catatonia [14]. Also recently published review of neuroimaging and functional neuroimaging data in catatonia are in line with this idea [15]. The authors also indicate important role of dopamine neurotransmission in cortical and subcortical areas, which seems to be associated with motor symptomatology [15].

Autonomic nervous system is another area of clinical and research interest in catatonia. Autonomic disturbances are common in catatonia and may lead to fatal outcome. Case study of experimental catatonia treatment by vagus nerve stimulation has been also published [16].

It is unclear why catatonia is so rare in children. Some researchers argue that catatonia is actually not so rare, it is just rarely properly diagnosed [13]. Nonetheless, by now this idea is not a common view.

Catatonia in children and adolescents

There are no separate criteria for catatonia in children and adolescents. However, in the previous decades catatonia in children was described and analyzed in the wider context than just schizophrenia and mood disorders [13].

Traditional conceptualization

Literature review focused on child catatonia has been recently presented by Benarous et al. [17]. Catatonia prevalence in the general population remains unknown. In child and adolescent inpatients prevalence ranged between 0.6 and 17 percent. Prepubertal catatonia was very rare. Catatonia was also associated with 60-fold increased risk of premature death in follow-up.

Schizophrenia is the most common disorder associated with catatonia in children. Catatonic schizophrenia was associated with more unfavorable course of the disorder in children and adolescents when compared to other types of schizophrenia [18].

Mood disorder is the second most common psychiatric disorder associated with catatonia in the population of children and adolescents, following schizophrenia spec-
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trum disorder. Mood disorders associated with catatonia include episodes of major depression and mania. The literature describes the case of cyclic catatonia in a 16-year-old boy with bipolar disorder [19]. The mechanism of this relationship is unknown.

Link between catatonia and experienced stress is still discussed. It is also unclear what is the association between catatonia and childhood-specific, trauma-related psychopathological syndromes, such as pervasive refusal syndrome with predominance of motor symptoms [17].

Another idea is discussed by Shorter and Wachtel [20, 21]. The authors after analysis of historical case studies emphasize the necessity of differentiation between schizophrenia and catatonia. On the other side, the hypothesis is proposed that the most typical form of child catatonia is a mixture of three by now separate diagnostic entities – catatonia, autism and psychosis.

Catatonia in autism spectrum disorder (ASD)

As it was mentioned above, according to the DSM-5 and ICD-11 classifications catatonia may be diagnosed as a specifier, e.g., in the course of autism spectrum disorder. This idea has been acknowledged in classification systems since 2013. Nevertheless, already in the year 2000 Wing and Shah [22] observed comorbid catatonia in 17% of autism spectrum disorder patients. In the studied population, all catatonia cases were in patients older than 15 years. Following studies [23, 24] confirmed that catatonia in the course of ASD may also be observed in children. Some researchers [25] consider catatonia one of ASD symptoms and propose the concept of “autistic catatonia”.

Differential diagnosis may indeed be a challenge in clinical practice – numerous symptoms linked with catatonia (e.g., stereotypies or echolalia) may also be present in autism.

Wing and Shah [22] described 4 main symptoms of catatonia in ASD:

(2) increasing psychomotor and verbal slowness;
(3) difficulties in initiating and completing actions;
(4) increased reliance on physical or verbal prompts;
(5) increased passivity and apparent lack of motivation

The same authors also noticed four frequently observed behavioral abnormalities occurring in patients with catatonia and ASD:

(1) reversal of day and night;
(2) Parkinsonian features;
(3) excitement and agitation;
(4) increase in repetitive, ritualistic behavior.

Breen and Hare [26] conducted studies in families of ASD patients and found that catatonic symptoms are more frequent in female than in male patients. Moreover,
the study showed worse functioning in patients with co-occurring ASD and catatonia symptoms – they score higher, when compared to individuals with no signs of catatonia, in the Attenuated Behavior Questionnaire (ABQ), which measures an increase in ASD symptoms severity in the course of illness.

The issue often raised in the discussion regarding link between ASD and catatonia is the trend of neglecting catatonia symptoms in autistic children. Moreover, some experts [27] postulate that every patient experiencing worsening of ASD symptoms should be screened for catatonia.

**Organic catatonia**

According to some experts, about 20% of catatonia cases in child and adolescent population has an organic background. The possible background includes genetic and metabolic disorders, infectious diseases (e.g., toxoplasmosis and viral encephalitis) autoimmune diseases, epilepsy, intoxication, and iatrogenic complications [28].

Metabolic and genetic disorders should be suspected in case of positive familial history and in case of exacerbation of catatonic symptoms associated with catabolism (e.g., fever or surgery). Another hint of possible organic catatonia etiology are sings of intra-tissue storage, for example, hepatomegaly [29]. Metabolic disorders associated with catatonia in the pediatric population include Wilson’s disease and porphyria [17].

Genetic disorders associated with catatonia include: Huntington’s disease, chromosomopathy or CNV mutations, such as Down syndrome, 22q13.3 deletion syndrome, Prader-Willi syndrome, fatal familial insomnia, Kleefstra syndrome, creatine deficiency, and Sanfilippo syndrome. Catatonia syndrome may also occur in the course of mitochondrial encephalomyopathy, for example, MELAS syndrome (mitochondrial encephalopathy, lactic acidosis, and stroke-like episodes) or mutations in the PRODH gene [30].

The autoimmune background may refer to systemic diseases, i.e., systemic lupus erythematosus as well as autoimmune encephalitis, e.g., anti-NMDA (anti-N-methyl-D-aspartate receptor) encephalitis or pediatric autoimmune psychiatric disorders associated with streptococcal infection (PANDAS) [31–33].

Catatonia associated with ecstasy, mephedrone, gamma-hydroxybutyric acid, mescaline, cocaine, and opiates intoxication, cannabis abuse or iatrogenic complication after: steroids, disulfiram, ciprofloxacin, baclofen, lithium, phencyclidine, chlorfenamine, ciclosporin, insulin, antiretroviral drugs, and bupropion treatment was also described [34].

**Autoimmune encephalitis**

Catatonic symptoms are relatively common in anti-NMDA (anti-N-methyl-D-aspartic acid) receptor encephalitis, which is a form of autoimmune encephalitis.
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There is a female predominance in this condition. From 45 to 60% of patients is 18 years old or younger. Anti-NMDA encephalitis is considered paraneoplastic syndrome commonly co-occurring with ovarian teratomas. In pediatric population tumors are less common [35, 36].

The first signs are often flu-like symptoms. Psychiatric symptoms, such as mood disorders, maniac or depressive episodes, anxiety, agitation, disorganized thinking, delusions, auditory and visual hallucinations, are present in more than 70% of patients with anti-NMDA encephalitis. Acute psychosis may be the cause of psychiatric admission. Approximately 70% of patients experience neurological dysfunction such as seizures and dyskinesia. Language deficits, memory impairment, confusion, autonomic instability (cardiac arrhythmias, hypotension, hypertension, dysthermia), and central respiratory dysfunctions are also frequent [35].

Differential diagnosis includes psychotic disorders, mood disorders, impulse control functional deficits, serotonin syndrome, neuroleptic malignant syndrome, intoxication, viral encephalitis, and other forms of encephalitis (e.g., lupus, antiphospholipid antibody syndrome) [35, 36]. Diagnosis of anti-NMDA encephalitis is based on clinical presentation and confirmed presence of NMDA receptor antibodies both in serum and cerebrospinal fluid [35]. Sometimes, but not always, magnetic resonance abnormalities are observed. Treatment is based on oncological surgery when needed and immunotherapy. Symptomatic and supportive care is also recommended, including typical catatonia management.

Catatonia in obsessive-compulsive disorder (OCD)

According to the newest versions of ICD and DSMA, catatonia may be diagnosed also in obsessive-compulsive disorder. A few case studies presenting patients with catatonia comorbid with OCD were published [37]. Most patients were adults, however, there is also a description of complex treatment in a patient in whom the 13-year-long catatonic syndrome caused by obsessive-compulsive disorder started at the age of 12 [38]. In all presented cases OCD symptoms preceded catatonic signs. Therefore, it seems that in adequate treatment planning not only addressing first-plane catatonic behaviors but also managing underlying symptoms seem to be a therapeutic challenge [37, 38].

Primary slowness is the sign considered by many researchers as one out of three basic OCD symptoms. In extreme severity primary slowness may take the form of catatonic-like behavior [39].
Outside ICD and DSM

Pervasive refusal syndrome (PRS)

Pervasive refusal syndrome is a condition described in the literature but not acknowledged in classification systems [40]. This rare disorder is considered to be associated with developmental age. Proposed criteria:

1. partial or complete refusal in three or more of the following domains: eating, mobilization, speech, social contacts;
2. active refusal to acts of help and encouragement;
3. social withdrawal and school refusal;
4. no organic or psychiatric condition accounts for the severity of the degree of symptoms
5. the patient’s condition is serious

Pervasive refusal syndrome may imitate catatonia, nevertheless psychogenic etiology is postulated. Association between trauma and PRS is suggested [40], for example, case series in refugee children in Sweden has been presented [41]. In the studied sample comorbid conditions included depression and post-traumatic stress disorder. The issue of differential diagnosis between stress-related conditions and PRS was not discussed in this study.

Prognosis in PRS is generally good. However, a case study of fatal outcome of PRS in the 11-years old girl was published in 2015 [42]. In the commentary to this work, another expert argue that actually the proper diagnosis in this case was not pervasive refusal syndrome but catatonia [43].

Lethal catatonia

This progressive, fatal form of catatonia was for the first time described by Calmeil in 1831 [as cited in: 44]. As it was summed up in the review published in 1986 [44], typical course includes about two-week prodromal period characterized by emotional lability, irritability and sleep and appetite disturbance. Next phase is lasting, severe agitation accompanied by disturbance of consciousness. At the same phase mutism, posturing and catalepsy were observed. Increasing behavior disorganization was parallel to growing somatic problems: fever, blood pressure lability, tachycardia. In analyzed cases, this phase was followed by somatic breakdown, fever, coma, cardiopulmonary insufficiency, and death. Attention was also paid to increased muscle tension and posturing. Considered etiological factors include somatic disorders like severe infections and encephalitis. Nevertheless, in most cases reviewed by Mann et al. [44] no underlying somatic condition was found. Single descriptions of lethal catatonia cases still appear in more recent literature [45]. There are also descriptions of somatic conditions with clinical manifestation taking a form of lethal catatonia [46].
Lethal catatonia and stress

In general, lethal catatonia has not been linked with acute stress reactions. However, recently a new idea was presented. The authors hypothesize that capture myopathy (sudden death of captured animals), takotsubo cardiomyopathy, excited delirium syndrome, and lethal catatonia have similar pathomechanism leading to fatal outcome. According to this hypothesis, stress in these conditions is the main etiological factor, while sensitization (for example, cocaine abuse in excited delirium syndrome) or mental disorder in catatonia are predisposing contributors [47]. The authors refer also to the paper by Moskovitz published in 2004 [48]. Moskovitz argues that catatonia is an evolutionary continuation of freeze response observed in terrified animals. According to the Moskovitz, benzodiazepines efficacy in catatonia may confirm this hypothesis.

Treatment of catatonia

Relatively high dose of benzodiazepines is a first-line treatment for catatonia [49]. In case of life-threatening condition, electroconvulsive therapy (ECT) is effective and safe in children and adolescents [17, 49, 50]. While the patient is treated for catatonic symptoms (symptomatic treatment), the underlying etiological cause must be looked for and treated without delay (etiological treatment) [51].

Benzodiazepines (BZD)

Despite the various causes of catatonia, the treatment procedure at the initial stage is standardized (symptomatic treatment). BZD are first-line treatment for catatonia and lorazepam is considered to be a first-choice drug for pediatric catatonia. In most cases, symptoms are drastically reduced within three hours after receiving 1 to 3 mg of lorazepam. When a positive response is observed, a titration should be completed to maintain the dose that achieves a complete resolution of symptoms [17].

In a naturalistic study of 66 children and adolescents with catatonia, it was found that benzodiazepines improved catatonia in 65% of cases. The mean daily dose of lorazepam was 5.35 ± 3.64 mg/day and reached 15 mg/day in some patients. There was no relation between dose and level of improvement, and side effects were few [49].

Lorazepam is generally well tolerated and does not cause sedation was seldom observed, especially when daily incremental doses are administered. When a higher dosage is needed, the patient should be monitored for excessive sedation, which tends to precede respiratory difficulties [52].
Electroconvulsive therapy (ECT)

If benzodiazepines at adequate doses are ineffective, ECT should be considered. In life-threatening situations like malignant catatonia or neuroleptic malignant syndrome the treatment should not be delayed [17, 52]. Systematic pretreatment and post-treatment evaluation, including symptom and cognitive assessment, is recommended. Case studies, retrospective chart reviews as well as prospective cohort studies confirm the benefit of ECT in children with catatonia [17, 49, 50]. Consoli et al. [50] analyzed 59 case studies of children and adolescents with catatonia treated with ECT. Response was favorable in 45 patients (76%), while partial improvement was noted in 3 patients (5%) and a lack of response in only one. The evolution of symptoms was not analyzed in the remaining studies.

In the sample with autoimmune conditions analyzed by Ferrafiat et al. [51] until immunosuppressive treatment was initiated all patients received high doses of BZD as first-line treatment. None of them required ECT.

Atypical antipsychotics

In addition to symptomatic treatment of catatonia, treatment of comorbid psychiatric condition should be provided. Costs and benefits of administration of antipsychotics in catatonia should be weighted for each individual case. The general notion, however, is to discontinue neuroleptics in the acute phase of catatonic episode because of their inefficacy and associated risk of exacerbation of catatonic symptoms [17, 52]. We must note here that Polish recommendations are not so unanimous [53]. Once treatment with benzodiazepines or ECT is started and catatonia improves, atypical antipsychotics may target psychotic symptoms, especially in patients with schizophrenia, or may be used as maintenance treatment in psychosis and mood disorders [17, 49, 52].

NMDA-receptor antagonists

Numerous articles reported the use of amantadine and memantine in catatonia in adults [54]. The vast majority of patients had schizophrenia spectrum disorders, and amantadine was primarily used as monotherapy, often with resolution after a few doses. Memantine was more commonly used in combination with benzodiazepines. The analyzed cases concerned mainly adults. Only few case reports focused on amantadine (100–500 mg three times a day) and memantine (5–20 mg/day) as a potential treatment for catatonia in youth [17].
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Etiological treatment

About 20% of juvenile catatonia is associated with organic condition [28]. Ferrafiat et al. [51] recently formulated the Causality Assessment Score (CAUS), which should be used in differential diagnosis and may be an aid in distinguishing organic and non-organic catatonia. The tool has been specifically prepared to analyze the cases of autoimmune versus non-organic catatonia.

In case of autoimmune catatonia immunosuppressive treatment should be started as quickly as possible [28, 51]. According to the literature, the first-line treatment includes corticoids, plasma exchange (PE) and intravenous immunoglobulins. In case of treatment resistance (30% to 40% of patients) or relapse, second-line treatment is to be considered. Immunosuppressive medications such as rituximab, cyclophosphamide and mycophenolate mofetil should be considered [49, 55]. In anti-NMDR encephalitis approximately 38% of patients had also a diagnosis of comorbid tumor. This should be taken into consideration during treatment planning and initiation [56].

Some organic conditions other than autoimmune conditions underlying catatonia have specific treatments as well. For example, some metabolic diseases may be treated with diet regimens, supplementation (creatine deficit), B12 vitamin and folates (deficit in methylenetetrahydrofolate reductase, MTHFR) or anti-storage drugs (Wilson’s disease) [29].

Supportive measures

In the course of catatonia, a broad range of complications may occur. Some patients require a high level of nursing care, adequate hydration by intravenous fluids, prevention of aspiration, nasogastric tube feeds or enteral feeding. Thrombophlebitis prophylaxis may also be considered [17, 52].

Literature search and limitations of the current review

First step during preparation of the study was analysis of diagnostic criteria in classifications ICD-10, ICD-11 and DSM-5. Warsaw Medical University database and PubMed database were searched. Search terms were: katatonia, podłoże, przegląd, autoimmuneologiczne, organiczne, dzieci i młodzież, catatonia, background, review, autoimmune, organic, catatonia, catatonia child, catatonia adolescent, catatonia history, catatonia and autism spectrum disorders, catatonia and obsessive-compulsive disorders. Papers in Polish available on-line were also searched, even if they were not indexed in PubMed. The authors aimed to present publications most reliable and relevant for clinical practice.

The current paper is not a systemic review, which is of course a limitation of the study. Nonetheless, aim of the paper was not only to present the newest data but also...
to review some ideas regarding previous and current catatonia conceptualization. That is the reason for not-systemic literature search.

Recapitulation

The catatonia conceptualization is still discussed despite numerous past and present studies. The idea of catatonia being a separate syndrome is now widely accepted and this is how the condition is defined in ICD-11 and DSM-5. Thus, previous mode of catatonia understanding may be questioned and should be reanalyzed. Numerous hypotheses regarding the catatonia nature has been proposed. Biological catatonia background is most often taken into consideration, nevertheless the role of (extreme?) trauma was also proposed.

Proper diagnosis and differential diagnosis seem to be crucial in clinical practice. Misdiagnosis may have fatal consequences. On the other hand, effective treatment in this condition is available.

References

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