Mini Review

iMedPub Journals
www.imedpub.com

Clinical Psychiatry
ISSN 2471-9854

2021
Vol.7 No.3:97

A Mini Review on Catatonia in Children and Adolescents

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Citation: Ridgeway L, Okoye A (2021) A Mini Review on Catatonia in Children and Adolescents. Clin Psychiatry Vol.7 No. 3:97.

Received date: April 26, 2021; **Accepted date:** May 10, 2021; **Published date:** May 17, 2021

Abstract

Catatonia is a serious condition that is potentially life threatening and can occur in children and adolescents. This short review highlights the importance of an awareness of the catatonia syndrome in both psychiatric and medical paediatric clinicians. In young patients, catatonia can be associated with various psychiatric, medical or developmental conditions. The timely identification of catatonia is essential as the treatment approach differs to that of the differential diagnoses and possible underlying conditions. The important aspects of a comprehensive clinical assessment, including the use of clinical rating scales, are outlined in this article. The pillars of medical treatment, firstly benzodiazepines and the second line treatment option of electroconvulsive therapy are discussed.

Keywords: Catatonia; Children; Adolescents

Introduction

Catatonia is a potentially life-threatening condition that is increasingly recognized in the paediatric population. The prompt identification and specific diagnosis of catatonia syndrome is critical, as there are well-defined and effective treatments available. Catatonia is characterized by motor and psychological symptoms that severely effect functioning. It is associated with numerous psychiatric, neurodevelopmental and medical disorders in children and adolescents. Catatonic symptoms can be continuous or fluctuant and variations in presentation can be broadly described as stuporous catatonia, excited catatonia, malignant catatonia and periodic catatonia [1-4]. Stuporous catatonia presents with psychomotor retardation, immobility, rigidity, staring and mutism [1-4]. In contrast, excited catatonia is characterised by prolonged periods of psychomotor agitation [1-4]. It can be possible to observe prolonged periods of symptom resolution in periodic catatonia [1-4]. Malignant catatonia is a severe form of the illness that is potentially life- threatening and consists of an exacerbation of catatonic symptoms and autonomic instability [1-4]. Although a prevalence rate for the general population is not available, the prevalence of paediatric catatonia in inpatient populations varies between studies, from 0.6% to 17% [1,2,5,6]. The majority of paediatric catatonia cases occur at pubertal ages [2,7]. In contrast to their adult counterparts, catatonia in children and adolescents is more common amongst males, with a male to female ratio of approximately 2:1 [1,2,8,9]. There is also a possible role for ethnic or cultural factors in the clinical expression of catatonia in youths [2], with a higher prevalence rate reported in an Indian sample [6].

Etiology

The etiology of paediatric catatonia can be broadly divided into three categories; psychiatric, medical and unknown. The most common psychiatric cause for catatonia in this age group is schizophrenia [1,2,10] followed by mood disorders [1,2,10]. Traumatic factors can also contribute to the precipitation of catatonia in youths, including, deprivation, abuse and trauma [10,11]. Neurodevelopmental disorders are associated with a higher risk of paediatric catatonia, including, Autistic Spectrum Disorder (ASD), Tourette's Syndrome, Down's Syndrome, childhood disintegrative disorders and Prader-Willi Syndrome [2,10,12]. An incidence rate of catatonia of 4-17% has been found in adolescents and adults with ASD, based on a review of 6 studies (n=811) in 2006 [13]. Clinician awareness of the association between catatonia and developmental disorders in young patients is vital as a diagnosis can be difficult in this group of patients due to the possible overlap of symptoms. In a case-control study of paediatric patients with catatonia from 1993-2009 (n=58), 31% were associated with a developmental disorder and 22% of cases were associated with a medical condition [7]. The medical conditions associated with paediatric catatonia include epileptic encephalopathy, infections (e.g. viral encephalitis, typhoid fever, toxoplasmosis), autoimmune diseases (e.g. neuro-psychiatric systemic lupus erythematosus and anti-NMDA receptor encephalitis, paediatric autoimmune neuropsychiatric disorders associated with streptococcal infections, encephalopathy associated with autoimmune thyroid disease), toxic-induced states (e.g. lithium, steroids, ecstasy) and many metabolic and genetic disorders [1,7,10,12,14]. Comorbid medical conditions require prompt investigation, diagnosis and appropriate treatment which can subsequently lead to an improvement in catatonic symptoms [7,14]. The diagnostic categories in the Diagnostic and Statistical Manual for Mental Disorders 5th edition (DSM-5) for catatonia reflect the above associations and are as follows; catatonia as a specifier for schizophrenic disorders (schizophrenia, schizoaffective disorder, schizophreniform disorder, brief psychotic disorder, substance-induced psychotic disorder), catatonia associated with another

mental disorder (e.g. Bipolar Affective Disorder, Major Depressive Disorder), catatonia due to a general medical condition and catatonia not otherwise specified (i.e. when no etiology is identified) [15]. The addition of the final category of catatonia not otherwise specified in the DSM-5 particularly aids the diagnosis of catatonia in young patients when an underlying condition may not be identifiable.

Discussion

Catatonia syndrome is generally homogenous in its presentation despite the diverse collection of associated psychiatric, developmental and medical conditions [1,7,14]. For all diagnostic categories of catatonia, the DSM-5 require the presence of three out of twelve of the following symptoms: stupor (i.e. no reactivity to the environment, no psychomotor activity), mutism (i.e. no or minimal verbal response), waxy flexibility (i.e. slight and even resistance to repositioning by the examiner), negativism (i.e. opposing or not responding to external stimuli or instruction), posturing (i.e. spontaneous and active maintenance of posture against gravity), catalepsy (i.e. passive induction of postures held against gravity), mannerisms (i.e. odd caricatures of ordinary actions), stereotypies (i.e. repetitive, frequent, non-goal directed movements), agitation, grimacing, echolalia (i.e. repeating words spoken by the examiner) or echopraxia (i.e. mimicking movements made by the examiner) [15]. Other symptoms include rigidity and automatic obedience. Catatonic symptoms are generally similar across ages [1,3,4,16]. Other symptoms that can be prominent in younger patients include repetitive movements, social withdrawal, refusal to eat or drink, and regressive symptoms, such as urinary incontinence [1,2,10,16]. Many of the patient groups discussed so far, are vulnerable to diagnostic overshadowing and the inappropriate attribution of catatonic symptoms to a comorbid disorder. Children and adolescents with ASD, schizophrenia and other developmental disorders may demonstrate motor symptoms, attributable to their disorder, that can mimic catatonic symptoms and complicate the assessment, including, stereotypies in ASD, soft motor signs in schizophrenia or extra-pyramidal symptoms due to antipsychotic treatment [5,16,12]. Diagnostic differentiation between catatonia and its comorbidities is important as treatment approaches differ and inadequate treatment could lead to a worsening clinical picture or malignant catatonia.

In order to ensure a comprehensive assessment of a young person with catatonic symptoms a multidisciplinary approach is essential [14]. Both an extensive psychiatric assessment and neurological assessment are necessary [14]. An equally important aspect of the assessment is a thorough medical work-up targeting the differential diagnoses discussed previously and this should include blood investigations with basic haematologic and metabolic measures, toxicology screen, brain imaging, autoimmune antibodies and other tests indicated by clinical examination [7,10,12,14]. Approaching the psychiatric diagnosis of catatonia, along with the DSM-5 diagnostic criteria, one can utilise clinical rating scales to assist in the diagnostic process and to estimate clinical severity. Taking into consideration the similar symptomatology in their adult counterparts, clinical rating scales that were originally designed for adults with catatonia have been employed in the assessment of paediatric catatonia, including the Bush-Francis Catatonia Rating Scale (BFCRS) [10,16]. The BFCRS consists of a 23-item scale which scores the presence and severity of catatonic symptoms from 0-3 [16]. The presence or absence of the first 14 items are utilized as a screening tool [16]. Subsequently, Cohen has developed a modified scale from the BFCRS specifically to assess children and adolescents, called the Paediatric Catatonia Rating Scale (PCRS) [16]. Along with the seventeen items included in the BFCRS, the following adjustments were made in the PCRS, withdrawal was elaborated further into refusal to eat or drink and social withdrawal; grimacing was combined with mannerisms; automatic obedience was replaced by automatic compulsive movements; and urinary incontinence, schizophasia and acrocyanosis were all added [16]. A lorazepam challenge test can be useful for diagnostic validation of catatonia, if clinical response is evident following a 1-2mg test dose of intravenous lorazepam [12]. However, it is important to note that a non-response to the lorazepam challenge test does not out-rule a diagnosis of catatonia, as demonstrated by a recent case described by the authors [17].

Catatonia in children and adolescents can present either acutely or through a more insidious process [2,9]. The duration of the condition can also vary from transient to chronic lasting weeks to months [9,10]. The akinetic nature of the condition can lead to medical complications, including, malnutrition, dehydration, pneumonia, pressure ulcers or venous thromboembolism [10,14]. The risk of these complications further emphasizes the value of multidisciplinary management [14]. There is also the risk of progression to malignant catatonia, characterised by autonomic dysfunction, fever, delirium, exacerbation of catatonic symptoms and an increased mortality rate [1,2,10,12]. The use of antipsychotic medication can increase the risk of progression to this severe form of catatonia [10,12].

Early recognition, specific syndromic treatment with benzodiazepines and appropriate medical work-up are fundamental in the effective management of catatonia in young people [7,10,12]. The importance of a catatonia diagnosis as well as the diagnosis of the possible underlying precipitating co-morbidity is emphasized by effective response to treatment irrespective of the etiology. Alongside the treatment of catatonic symptoms, it is important not to neglect the appropriate treatment of the associated psychiatric or medical condition if present. A young patient with catatonia will require hospitalization and a multidisciplinary approach for assessment and treatment [3,14.] The general principles of the symptomatic treatment of catatonia in youths do not differ greatly from adults [4]. Similarly, first line treatment in children and adolescents is with benzodiazepines, most frequently lorazepam [3,9,10,12]. Fundamentally, a reluctance to prescribe benzodiazepines in young patients should not prevent their use in paediatric catatonia [3]. A lorazepam challenge test assists in the validation of the diagnosis and the assessment of response to treatment [10,12]. If a clinical response is evident, the lorazepam dose should be titrated for optimal response and maintenance of improvement [3,10,12]. It is important to note

that an absence of a response to the lorazepam challenge test does not out rule a diagnosis of catatonia [17]. In a prospective cohort study (n=66), benzodiazepine treatment was effective in 65% of patients with catatonia, aged 9-19 years, and the most frequently used benzodiazepine was lorazepam [3]. Severe adverse events associated with benzodiazepines were reported in 12% and included leukopenia, excessive sedation, agitation and one respiratory distress episode [3].

The second treatment option for catatonia in children and adolescents is electroconvulsive therapy (ECT). It is the next available treatment if the catatonic symptoms are refractory to optimal benzodiazepine titration. In a comprehensive review of the literature (n=396), an overall efficacy rate of 80% was reported for the use of ECT in children and adolescents with catatonia along with a similar side-effect profile with ECT in adults [18]. A more recent review of the literature demonstrated a concordant response rate to ECT in paediatric catatonia of 86% [19]. The American Academy of Child and Adolescent Psychiatry has issued practice parameters on ECT use in children and adolescents to help provide guidance to treating psychiatrists [20]. However, despite ECT being an effective treatment, there are many barriers to its use in the paediatric population. For example, legal barriers to ECT access for children and adolescents, the existence of resistance to ECT in both the medical and lay communities and the overall stigma attached to the treatment [21].

Conclusion

In conclusion, it is important for all paediatric clinicians, both psychiatric and medical, to have an awareness of catatonia syndrome to allow for early recognition, appropriate treatment and subsequent better outcomes for these young patients with a potentially life-threatening illness. The DSM-5 criteria can be used to guide diagnosis with the assistance of clinical rating scales. In young patients it is essential to carefully investigate for possible underlying medical causes that may also require treatment. Underlying psychiatric or neurodevelopmental disorders should also be identified. A multidisciplinary approach to treatment is beneficial. The primary treatment approach is benzodiazepine treatment with a second-line treatment option of electroconvulsive therapy.

Declaration of conflicts of interest

The authors do not have any conflicts of interest to declare.

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