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Catatonia in children and adolescents: New perspectives

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ABSTRACT

Introduction: Catatonia is a rare and severe psychomotor condition in children and adolescents. In the current report, we aimed to review the recent literature.
Method: Using a PRISMA approach, we searched MEDLINE between 1982 and 2017 using the keywords 'CATATONIA' and 'CHILD' or 'ADOLESCENT'. In total, we reviewed 130 reports (controlled study, $N = 4$; clinical chart, $N = 23$; case report, $N = 54$; and editorial/review, $N = 42$).
Results: Several aspects seem to be age specific: (1) although the clinical presentation resembles that in adults, some symptoms are important in children and adolescents (e.g., psychomotor regression). (2) Associated disorders are similar to that found in adults; however, schizophrenia is more frequently observed than mood disorder. Additionally, a history of neurodevelopmental disorders maybe encountered. (3) Morbidity and mortality are among the worst in child psychiatry. (4) Underlying organic conditions are highly prevalent (>20% of the cases), and their search is warranted because some diagnoses may result in specific treatments (e.g., immune-suppressor therapy for autoimmune conditions). (5) Symptomatic approaches – high dose of benzodiazepines and electroconvulsive therapy (ECT) – are as efficient in children or adolescents as they are in adults, but this finding needs to be acknowledged because a resistance against the use of ECT or high-dose medication exists among child psychiatrists.
Discussion: Recent advances in child and adolescent catatonia research have offered major improvements in understanding catatonia and in new therapeutic opportunities. The syndrome is rare, but these advances need to be acknowledged in order to direct patients to centers that have developed a specific expertise.

1. Introduction

While catatonia has been described as an adult condition, catatonic symptoms have been reported in children or adolescents since the nineteenth century. In a series of 26 adults with catatonia, Kahlbaum noted that the majority had their first symptoms in childhood (Kahlbaum, 1874). Raecke (1909), who presented the first clinical series in youths ($n = 10$), observed that the presentation was comparable between children and adults. The first attempt to separate catatonia from other mental conditions in children was made by Karl Leonhard (1979), who listed the differences between "infant catatonia", autism and the "state of feeblemindedness" (Leonhard, 1979).

Leonhard's research on youths with neuro-developmental disorders helped distinguish catatonia from motor dysfunctions associated with autism (Ohta et al., 2006; Wachtel and Dhossche, 2010; Wing and Shah, 2000). In the same vein, the observations made by Cohen et al. (1999) and Dhossche et al. (2006) in cohorts of inpatient youths promoted a syndromic view of the condition. This perspective, which has progressively been internationally endorsed (American Psychiatric Association, 2000, 2013), has also contributed to the acceleration of evidence-based research development and helped in the recognition of catatonia in children and adolescents.

In this article, we provide a review on catatonia in children and adolescents. Section 3 presents the epidemiology and the phenomenology of the syndrome, including the differential diagnoses. Section 4 summarizes the etiological factors and disorders associated with catatonia in children and adolescents. Section 5 attempts to propose a comprehensive model for catatonia. Finally, Section 6 provides an overview of therapeutic approaches.

2. Methods

The systematic review was conducted following the recommendations outlined in the PRISMA guide (Moher et al., 2009). To take into account relevant papers that were written in English, MEDLINE databases between 1982 and 2017 were searched using key terms that included

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'CATATONIA' and 'CHILD' or 'ADOLESCENT' in addition to manual searches. Titles and abstracts were scanned for relevance. Full texts were ordered in case of uncertainty to maximize sensitivity. Reference lists of retrieved systematic reviews were checked. All full texts were checked for eligibility. Studies in which the onset age of every subject with catatonia was over 18 and those that did not specify the onset age as either over or under18 were excluded. Any controlled study, clinical chart, case report, review or editorial were eligible for inclusion in this review. Of the 130 studies obtained by this method, 4 were controlled studies, 23 were chart reviews without case presentations, 54 were case reports, and 42 were editorial/review articles on a specific issue. Seven articles were beyond the scope of this article, and thus, they were excluded (Fig. 1, for detailed research strategy see Table S1). In addition to the report of current evidence on pediatric catatonia, we also mention possible clinical strategies and research prospects that deserve more attention based on our experience in treating youths with catatonia.

3. Phenomenology and diagnosis of catatonia in children and adolescents

3.1. Epidemiology

A prevalence rate for the general population is not available, which indicates that catatonia is a rare clinical syndrome in children and adolescents. The prevalence of catatonia in inpatient youths varies from 0.6% to 17% (Cohen et al., 2005; Takaoka and Takata, 2003; Thakur et al., 2003; Wing and Shah, 2000). In the overwhelming majority of cases, catatonic episodes occur in patients at pubertal ages (Consoli et al., 2012) and exceptionally at pre-pubertal ages (e.g., Wachtel et al., 2008). Furthermore, although the phenomenology of catatonia in young people is similar to that reported in the adult literature (see below), the sex ratio is different, with more boys affected than girls (sex ratio approximately 2:1) (Cohen et al., 1999; Takaoka and Takata, 2003).

3.2. Clinical description and diagnosis

Catatonia is a syndrome of abnormal motor function. Catatonic symptoms can be classified into motor (e.g., posturing, catalepsy, waxy flexibility), behavioral (e.g., negativism, mutism), affective (e.g., uncontrollable emotional reactions, withdrawal), and regressive symptoms (e.g., enuresis). Isomorphism across ages is supported by empirical studies (Dhossche et al., 2010) and has been adopted in the international classification. The DSM-5 criteria for catatonia include the presence of three symptoms from the following list of twelve: stupor, catalepsy, waxy flexibility, mutism, negativism, posturing, mannerisms, stereotypy, agitation, grimacing, echolalia and echopraxia (American Psychiatric Association, 2013) (Table 1). Other common symptoms are motor resistance to simple commands, posturing, rigidity, automatic obedience, and repetitive movements. While the DSM-IV uses different sets of criteria for the diagnosis of catatonia in schizophrenia and primary mood disorders versus neurological/medical conditions, the DSM-5 has adopted the syndromic approach promoted by most catatonia experts (Dhossche et al., 2010; Francis et al., 2010). In our own cohort, contributions to the classification of catatonia symptoms were limited, and no symptoms appeared pathognomonic of any psychiatric diagnoses, neuro-developmental histories or organic conditions (Benarous et al., 2016; Consoli et al., 2012). In addition to the DSM classification, we previously proposed a specific classification of catatonia in children and adolescents, as described in Table 2 (Cohen, 2006). This assumption was empirically derived from studies on the phenomenology of catatonia in youths (Benarous et al., 2016), the course of the disorder (Cornic et al., 2009), and its association with psychiatric and organic conditions (Consoli et al., 2012; Lahutte et al., 2008).

3.3. Clinical scales

One of the challenges in using rating scales is that catatonic symptoms fluctuate over time, and a longer period of observation may be required to obtain the full clinical picture. Several rating scales that were initially developed for adults were used in pediatric patients, such as

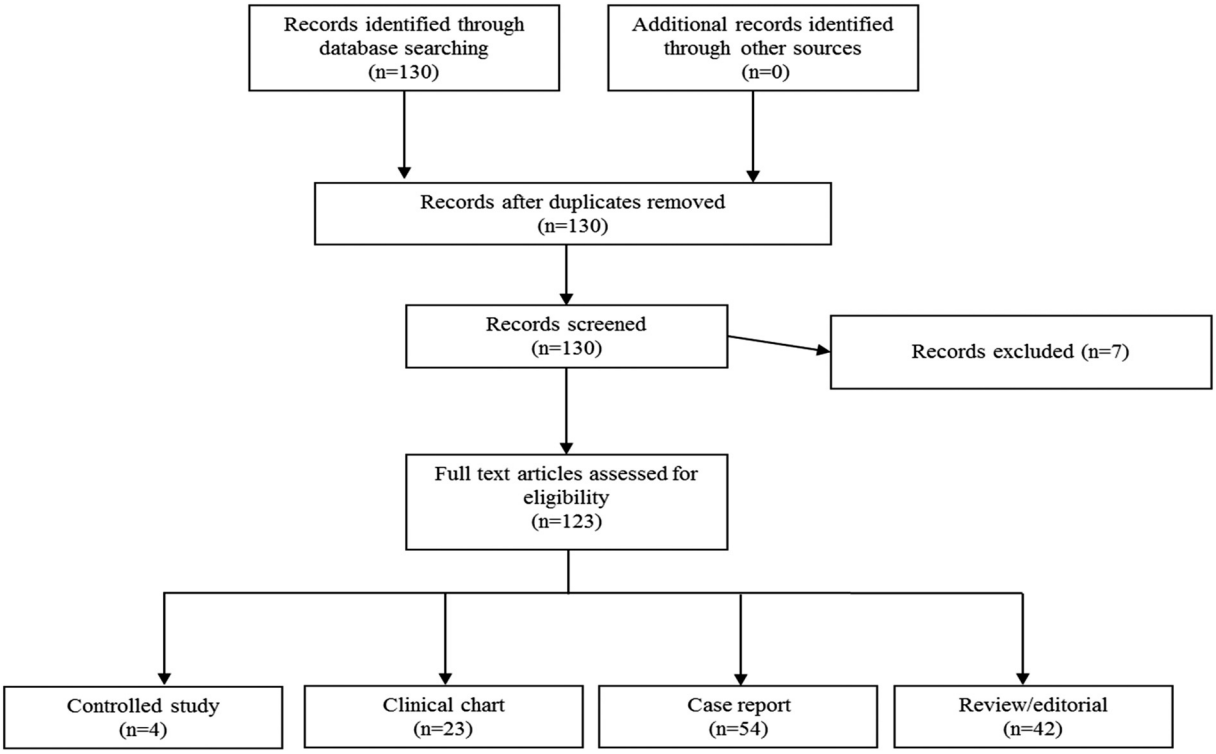

Fig. 1. PRISMA diagram flow of the study search.

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