Validation of the Pediatric Catatonia Rating Scale (PCRS)

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A B S T R A C T

Introduction: Despite the increased recognition of catatonia in children and adolescents, no specific assessment instrument has been validated in this population.
Method: Within the context of a prospective study on catatonia, we developed the Pediatric Catatonia Rating Scale (PCRS, maximum score = 60), adapted from the Bush and Francis Catatonia Rating Scale for its use in child and adolescent inpatients. We assessed the psychometric properties of the PCRS by measuring its internal consistency, construct validity, and factor structure. Bivariate analyses were performed to compare the different diagnostic patient groups across the extracted factors.
Results: Internal consistency was moderate (Cronbach's α for total score = 0.67) suggesting multidimensionality. Multiple factors underlie the PCRS items, as revealed by factor analysis. Four distinct dimensions of catatonic symptoms were identified and accounted for 44% of total variance: a "negative withdrawal" factor (with mutism, negativism, and social withdrawal), a "catalepsy" factor (with posturing and waxy flexibility), an "abnormal movements" factor (with mannerisms and stereotypes) and an "echo phenomenon" factor (with echolalia and echopraxia). Receiver operating characteristic (ROC) analysis showed that the PCRS performance in discriminating individuals with catatonia vs. those without catatonia was excellent for a threshold ≥ 9 (Area Under the Curve = 0.983) in this sample.
Discussion: These results support the validity of the PCRS among children and adolescent inpatients. With regard to such analyses, the internal structure of catatonic syndrome in children and adolescents is roughly comparable with the adult form, except the lack of a "hyperactive/excitement" dimension.

1. Introduction

Pediatric catatonia has attracted considerable interest in recent decades (Dhossche et al., 2010; Taylor and Fink, 2003). The possibility that motor symptoms (e.g., mutism, negativism, or staring) in children or adolescents may constitute a distinct syndrome has substantial prognostic and therapeutic implications (Cohen et al., 1999). Indeed, pediatric catatonia is a serious disease with possible lethal consequences in its severe form (i.e., malign catatonia) (Cornic et al., 2009), for which treatments are easily available and effective in most cases (Raffin et al., 2015). The idea that catatonia is a transnosological syndrome was supported by prior studies in adults showing that treatments are effective regardless of the etiology (Bush et al., 1996a), and that symptoms

involved common pathophysiological mechanisms (Abrams et al., 1979; Northoff et al., 1999; Taylor and Fink, 2003). Catatonia is not classified as a separate disorder under DSM-5, it is instead a state associated with a number of disorders including mood (i.e., depression and mania) and psychotic disorders (i.e., schizophrenia, brief psychotic disorder, schizophreniform disorder, schizoaffective disorder, and substance-induced psychotic disorder) (American Psychiatric Association, 2013). In addition, the categories "catatonia due to a general medical condition" and "catatonia - Not Otherwise Specified [NOS]" (i.e., when no etiology is identified) are set as distinct disorders in the DSM-5. In this study, the terms "catatonic syndrome" and "catatonic episode" are regarded as similar and encompasses the disorders listed above.

The prevalence of catatonia in inpatient youths varies by a factor of thirty between studies (from 0.6% to 17%) (Cohen et al., 1999), and the possibility this syndrome could be underestimated in the pediatric clinical setting was raised on the basis of the findings of a systematic clinical assessment (Thakur et al., 2003). Among other reasons, a lack

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of consensus regarding the definition of a catatonic episode at this age could be an obstacle to a timely diagnosis (Cohen et al., 1999; Wong et al., 2007). Although case reports highlighted the developmental specificities of catatonia in youths, isomorphism across ages was supported by empirical studies (Dhossche et al., 2010) and adopted in the international classification (American Psychiatric Association, 2013). In this vein, almost all of the studies conducted used measurement tools originally developed for adults without any further adaptations. In particular, the Bush–Francis Catatonia Rating Scale (BFCRS) was the first and most widely used instrument constructed for the standardized and quantifiable examination of catatonia (Bush et al., 1996a). Although the validity of the BFCRS was demonstrated against the clinical gold standard (Bush et al., 1996a) by predicting the treatment response to lorazepam (Bush et al., 1996b) in adults, its psychometric properties have never been examined in a pediatric sample. The development of a valid assessment instrument for catatonia in youths would be an important step toward the better management of this syndrome.

Substantial progress has been made in delineating catatonia across a wide range of psychiatric disorders in children and adolescents (Cohen et al., 2005; Consoli et al., 2012; Dhossche et al., 2010; Dhossche, 2004; Takaoka and Takata, 2003; Taylor and Fink, 2003; Thakur et al., 2003; Wing and Shah, 2000). Unlike adults, the most common underlying psychiatric disorder is schizophrenia (Cohen et al., 2005; Takaoka and Takata, 2003), and catatonic syndrome can also occur in youths with a history of developmental disorder (e.g., autistic spectrum disorder [ASD] or intellectual disability [ID]) (Dhossche, 2004; Wing and Shah, 2000). In addition to various psychiatric disorders, catatonia may also occur in patients with medical conditions (e.g., neurological, auto-immune, and metabolic diseases) (Consoli et al., 2012; Lahutte et al., 2008) or result from intoxication (Masi et al., 2002; Maxwell et al., 1993). Difficulties in identifying catatonia at this age stem from the possible overlap between catatonic symptoms and symptoms of comorbid disorders (Dhossche et al., 2010). Youths with schizophrenia, ASD, and other developmental disorders may exhibit persistent abnormal motor symptoms that result from a clinical expression of the disorder (e.g., stereotypies in youths with ASD), motor neurological soft signs (e.g., in children with early-onset schizophrenia), or extrapyramidal syndrome linked to antipsychotic pharmacotherapy (McKenna et al., 1991; Raffin et al., 2015; Wing and Shah, 2000). Such signs characterized by aberrant motor functioning can mimic catatonic symptoms and complicate clinical assessment (Dhossche, 2004; Wong et al., 2007); particularly in youths where schizophrenia and developmental disorders are found at higher rates in patients with catatonic episodes than in their adult counterparts (Consoli et al., 2012). From a clinical perspective, the distinction between catatonic symptoms and other motor symptoms is important as the therapeutic strategies may differ between the two situations and an inadequate treatment may worsen clinical signs and lead to malign catatonia. Then, comparing catatonic symptoms across psychiatric disorders should provide useful insight into the key symptoms of catatonia in youths that lead to a better identification. From a research perspective, such a comparison helps to test the stability of the structure of catatonic symptoms across psychiatric disorders.

Cohen et al. (2005) used a modified version of the BFCRS to study catatonic syndrome in child and adolescent inpatients. It has been used to explore the phenomenology of catatonic syndrome in youths (Cohen et al., 1999), to compare the presentation across different etiologies (Cohen et al., 2005), to follow the course of symptoms (Cornic et al., 2009), and to measure treatment response (Raffin et al., 2015). The scale has not been developed as a screening instrument for catatonia in clinical settings. Changes from the original scale were made based on a review of historical clinical studies (Ey, 1950), and were empirically derived from a comparison of the frequencies of catatonic symptoms across age groups (i.e., an analysis of 463 catatonic cases pooled from seven studies) (Cohen et al., 1999). Symptoms taken from Ey's earlier

description were added (Ey, 1950) (i.e., incontinence, acrocyanosis, schizophasia, automatic compulsive movements) and withdrawal was separated into refusal to eat/drink and social withdrawal (Cohen et al., 2005). This modified scale was called the Pediatric Catatonic Rating Scale (PCRS).

The aims of this study were to determine the reliability and validity of the PCRS in inpatient children and adolescents and to examine whether catatonic symptoms vary among diagnostic groups.

2. Methods

2.1. Catatonia ample

Every child or adolescent inpatient admitted to the Department of Child and Adolescent Psychiatry at University Hospital La Pitié-Salpêtrière was systematically assessed for catatonic symptoms between 1993 and 2015. During the time period of the study, 6463 patients aged 4–18 years were hospitalized. The screening for catatonic syndrome follows a two-step procedure. First, at entry or during the course of hospitalization, each patient with a catatonic motor sign was examined by one of the senior psychiatrists in charge of the study. Regarding catatonic motor symptoms, most of the patients were referred because of extrapyramidal symptoms secondary to antipsychotic prescription and were not eligible. Second, the diagnosis of catatonic syndrome was made by a senior psychiatrist and subjects were included for full clinical assessment during the period of hospitalization.

Criteria for the diagnosis of pediatric catatonic syndrome follow previous recommendations in literature in view of facilitating the identification of catatonic syndrome in youths while preventing over diagnosis (Cohen, 2006; Dhossche, 2014; Thakur et al., 2003; Wing and Shah, 2000). In particular, two points were highlighted in previous works. First, in youths with developmental disorders, a catatonic episode can be diagnosed only if a sharp and sustained increase of symptoms lasting days or weeks is observed or elicited (Dhossche, 2014; Wing and Shah, 2000). Second, non-motor catatonic symptom (i.e., behavioral/emotional/autonomic), as originally defined in the seminal study of Bush et al., (1996a), are particularly worth investigating during clinical assessment in youths considering the low specificity of abnormal motor symptoms at these age (Cohen, 2006). In line with Cohen (2006), the diagnosis of catatonic syndrome was made in the presence of at least two abnormal motor symptoms, or one motor symptom combined with a non-motor symptom (details are provided in Table 1). The full version of the PCRS is presented in Appendix A.

2.2. Control sample

To ensure that PCRS was more specific to catatonic symptoms than other motor symptoms that can be encountered in young psychiatric patients, we also recruited a control sample to perform a receiver operating characteristic (ROC) analysis. We chose a control group that was particularly enriched in medicated youths with antipsychotic drugs. Considering the very high sensitivity to neurological adverse events observed in youths treated with antipsychotics (Raffin et al., 2015), a point of particular relevance is to determine whether medicated youths do not score high to the PCRS.

The control group was selected as follow. First we included all the patients from our site participating in the ETAPE study [Trial registration number: NCT02007928 (http://www.clinicalstrials.gov)], which investigates the incidence of side effects of antipsychotics in youths (Menard et al., 2014). The ETAPE study is an ongoing naturalist multicenter study conducted over a 3 year follow-up. Enrolments started in 2013. Six to 18-year-old inpatient subjects who were given an antipsychotic treatment for the first time (or never more than three months

Table 1
Comparison between different diagnostic criteria for a pediatric catatonic episode.

Description of items of catatonia	DSM-5 criteria	BFCRS items	PCRS items
Catalepsy (i.e., passive induction of a posture held against gravity)	X	**X**	X
Waxy flexibility (i.e., slight and even resistance to positioning by examiner)	X	**X**	X
Stupor (i.e., no psychomotor activity; not actively relating to environment)	X	**X**	X
Agitation (i.e., not influenced by external stimuli)	X		
Mutism (i.e., no, or very little, verbal response, not applicable if there is an established aphasia)	X	**X**	X
Negativism (i.e., opposing or not responding to instructions or external stimuli)	X	**X**	X
Posturing (i.e., spontaneous and active maintenance of a posture against gravity)	X	**X**	X
Mannerisms (i.e., odd caricature of normal actions)	X	**X**	X
Stereotypes (i.e., repetitive, abnormally frequent, non-goal directed movements)	X	**X**	X
Grimacing (i.e., maintenance of odd facial expressions)	X	**X**	
Echolalia (i.e., mimicking another's speech)	X	**X**	X
Echopraxia (i.e., mimicking another's movements)	X	**X**	X
Excitement (i.e., extreme hyperactivity, constant motor unrest which is apparently non purposeful. Not to be attributed to akathisia or goal directed agitation)		**X**	X
Staring (i.e., fixed gaze, little or visual scanning of environment, decreased blinking)		**X**	X
Rigidity (i.e., maintenance of a rigid position despite efforts to be moved, exclude if cogwheeling or tremor present)		**X**	X
Verbigeration (i.e., repetition of phrases or sentences, like a scratched record)		**X**	X
Withdrawal (i.e., refusal to eat, drink and/or make eye contact)[a]		**X**	X
Impulsivity (i.e., patient suddenly engages in inappropriate behavior without provocation. Afterwards can give no, or only facile explanation)		**X**	
Mitgehen (i.e., "anglepoise lamp", arm raising in response to light pressure or finger, despite instruction to the contrary)		**X**	
Ambitendency (i.e., patient appears motorically "stuck" in indecisive hesitant movement)		**X**	
Perseveration (i.e., repeatedly returns to same topic or persists with movement)		**X**	
Autonomic abnormality (i.e., circle temperature, BP, pulse, respiratory rate, diaphoresis)		**X**	X
Automatic obedience (i.e., exaggerated cooperation with examiner's request or spontaneous continuation of movement requested)		**X**	
Gegenhalten (i.e., resistance to passive movement which is proportional to strength of the stimulus, appears automatic rather than willful)		**X**	
Grasp reflex (i.e., per neurological exam)		**X**	
Combativeness (i.e., usually in an undirected manner, with no, or only a facile explanation afterwards)		**X**	
Incontinence (i.e., nocturnal enuresis, daytime urinary incontinence, or fecal incontinence)[b]			X
Automatic compulsive movements (i.e., involuntary muscle activity exhibited in posture, attitudes, mimic or gesture due to inhibition or forced motor action)[c]			X
Schizophasia (i.e. scrambled speech)[d]			X
Acrocyanosis (i.e., cyanosis of the extremities)[e]			X

Diagnosis of pediatric catatonia

DSM-5 (APA 2013): 12 symptoms. Subjects have to present a minimum of three of the 12 catatonic symptoms. DSM-5 criteria were not available at the time of the studies.

BFCRS (Bush et al., 1996a): 14 items for screening (in bold), and a total of 23 items to rate severity. The scale encompasses 30 catatonic symptoms. Compared to DSM diagnosis criteria some of them are merged (e.g., Posturing/Catalepsy, Echopraxia/Echolalia, and Immobility/Stupor).

PCRS: 20 items. In addition to the 14-item of the BFCRS, the following six symptoms were added based on the analysis of 463 catatonic cases pooled from seven studies and review of historical description of pediatric catatonia (Cohen et al. 2006): (a) Withdrawal was separated into refusal to eat/drink and social withdrawal to ease distinction with other childhood psychiatric disorders, (b) Incontinence, a symptom of general psychomotor regression frequently reported (in 45% of pediatric catatonia in Dhossche and Bouman's series 1997), (c) Automatic compulsive movements encompass Grimacing and was regarded as an expression of psychomotor automatism (De Clérambault, 1927) it would predict a higher risk of presenting an underlying psychotic disorder, (d) Schizophasia, and (e) Acrocyanosis was added an expression of malignant catatonia in youths in addition to the item "Autonomic abnormality".

Similarly to BFCRS, each symptom was rated on a scale from 0 (absent) to 3 (severe), leading to a maximum score of 60. Subjects has to present at least two motor symptoms of a least 1 h duration or which can be observed or elicited on two or more occasions. Alternatively, subjects may exhibit one motor symptom and one non-motor catatonic symptoms. Distinction between motor and non-motor symptoms was developed by Bush et al. (1996) and further operationalized to facilitate identification of catatonia in pediatric sample by Cohen et al. (1999).

Motor symptoms: Catalepsy, Waxy flexibility, Stupor, Negativism, Posturing, Stereotypies, Excitement, Staring, Rigidity, Automatic compulsive movements,

Non-motor symptoms (i.e., behavioral/emotional/autonomic): Mutism, Mannerism, Echolaly, Echopraxia, Verbigeration, Withdrawal, Autonomic abnormality, Incontinence, Schizophasia, Acrocyanosis

successively) were included. A full clinical assessment was conducted to identify the main psychiatric diagnoses. The study protocol included that the PCRS was systematically administered to all subjects in view of estimating the possible impact of a first antipsychotic treatment on the emergence or the exacerbation of catatonic symptoms. From the initial sample of youths already included in the Pitié Salpêtrière Hospital center (n = 43), 3 youths diagnosed with catatonia were excluded. Only the PCRS rated at 6 months was used to ensure a certain time of antipsychotics exposure (n = 40).

Second, we decided to also include a small sample of antipsychotics-free inpatient subjects in the control group (n = 10). There was no other limitation regarding past or current medication. These subjects were all hospitalized in the Child and Adolescent Psychiatric Department at the Pitié Salpêtrière Hospital between November and December 2014 in the intensive psychiatric care unit. No exclusion criteria were applied to provide a representative picture of inpatients population. The study was conducted according to the hospital ethics committee's regulations. Fig. 1 summarizes the diagram flow of the study participants.

2.3. Measures

The 20 items of the PCRS were completed by senior psychiatrists who had experience in assessing catatonia and were trained in the use of the PCRS. All items were completed for 86 individuals (98% of the sample). A hot-deck imputation procedure was used to handle missing data. Other measures included the Clinical Global Impression-Severity scale (CGI-S), the Clinical Global Impression-Improvement scale (CGI-I) and the Global Assessment Functioning scale (GAF) which were administered upon admission and discharge. The Diagnostic Interview for Genetic Studies (DIGS) version 2.0, a semi-structured diagnostic interview developed by the Human Genetics Initiative of the National Institute of Mental Health, assessed lifetime and current DSM-IV psychiatric diagnoses (www.nimhgenetics.org). Information regarding
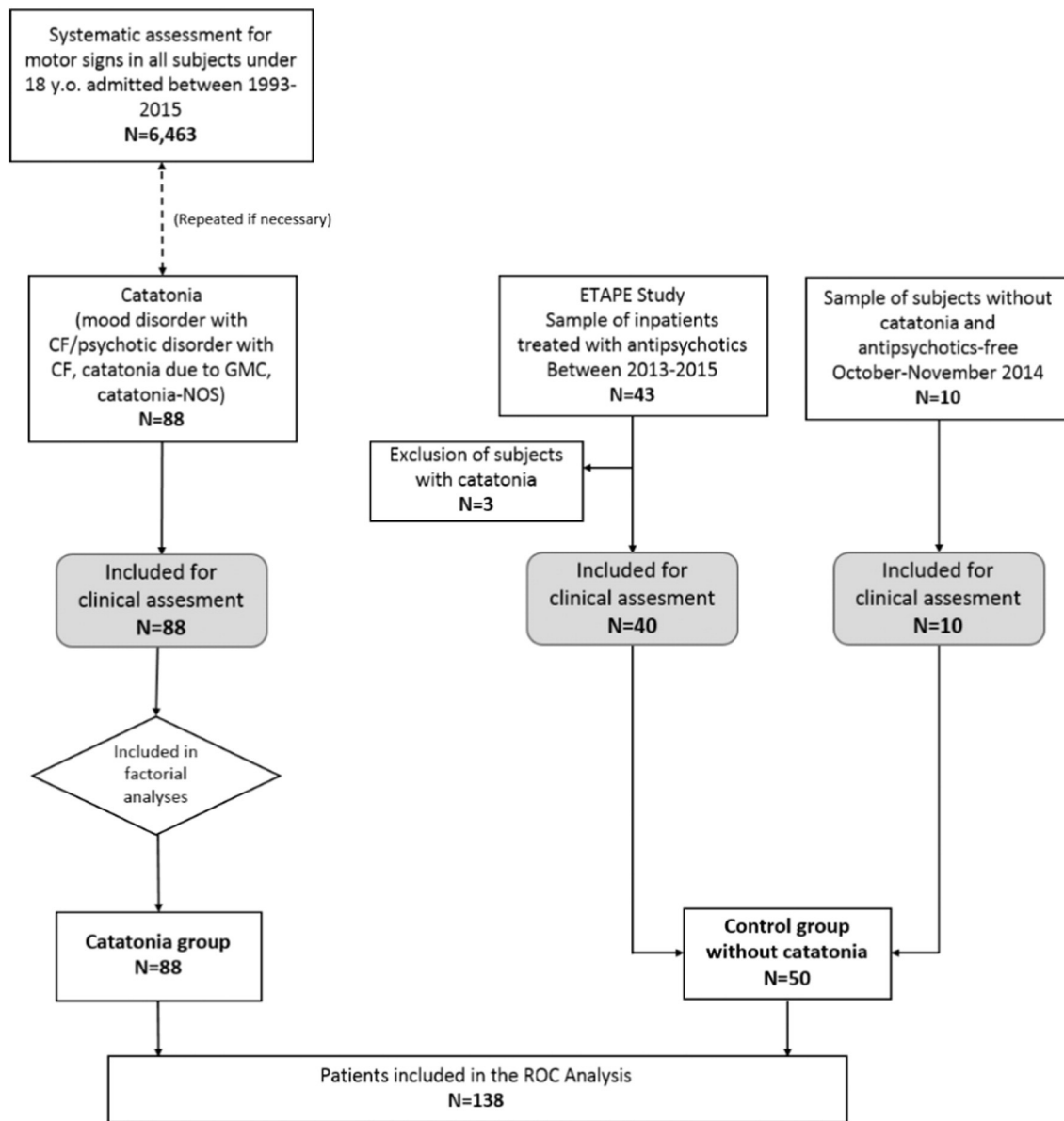
Fig. 1. Flow chart. *Note*: CF: catatonic features; GMC: General Medical Condition; NOS: Not Otherwise Specified; MS: Motor Symptoms; NMS: Non-Motor Symptoms. Number of subjects excluded due to the presence of extrapyramidal symptoms was not reported.

the type of catatonia onset (i.e., <10 days = acute; >10 days = insidious) and the duration of the episode (i.e., <6 months = episodic or > 6 months = chronic) were documented.

2.4. Statistical analyses

Data analysis proceeded in five steps. First, reliability was investigated with respect to internal consistency (Cronbach's alpha). Second, construct validity was examined by calculating Pearson's correlation coefficients between the PCRS total score and other measures. Positive correlations between the PCRS score and the duration of hospitalization, as well as a measure of global functioning (i.e., GAF) would support convergent validity; while divergent validity would be endorsed if no significant association was found between the PCRS score and a measure of stressful life events, the Adverse Childhood Experience (ACE) score. Third, an exploratory factor analysis was performed to explore the internal structure of the PCRS. To identify the most parsimonious number of dimensions,

a scree plot was generated and parallel analysis was performed. The Tucker Lewis Index (TLI) of factoring reliability, the Root Mean Square Error of Approximation (RMSEA) index and the Velicer Minimum Average Partial (MAP) criterion were examined to determine the suitability of data for factor analysis. As the PCRS uses a 4-point Likert scale, the estimation method was the weighted least-squares method (Muthén and Kaplan, 1985). Considering the moderate correlation between factors, a promax rotation was performed (Gorsuch, 1983). Any item loading >0.40 was considered to be significant. Fourth, bivariate analyses were performed to assess group differences in PCRS scores and in the distribution pattern of catatonic symptoms across diagnoses at discharge. The dimensions of catatonic symptoms were compared between the subjects diagnosed with affective disorders and those with schizophrenic disorders and then between youths with and without developmental disorders (i.e., intellectual disability and/or autistic spectrum disorder). Subscales were built by summing the items associated with each factor. Differences between patient groups were assessed

Table 2
Demographic and clinical characteristics of children and adolescents with catatonia.

	Catatonic subjects (n = 88)	Non catatonic subjects (n = 50)
Socio-demographic characteristics		
Gender, female, n (%)	31 (35%)	8 (16%)
Age (y) (mean ± SD)	15.17 ± 2.95	12.91 ± 2.78
Socio-economic status, high and middle, n (%)	60 (68%)	38 (76%)
Psychiatric diagnoses		
Schizophrenia	48 (55%)	4 (8%)
Major depressive episode	33 (38%)	15 (30%)
Manic episode	5 (6%)	3 (6%)
Autistic spectrum disorder	22 (25%)	7 (14%)
Intellectual disability	15 (17%)	4 (8%)
Other psychiatric diagnoses	0 (0%)	20 (40%)
Clinical characteristics		
Onset (sudden, ≤10 days), n (%)	36 (41%)	–
Duration (acute), n (%)	43 (49%)	–
GAF admission (mean ± SD)	19.26 ± 8.55	46.33 ± 12.98
GAF discharge (mean ± SD)	51.73 ± 14.69	NA
CGI-S (mean ± SD)	6.74 ± 0.45	4.40 ± 1.01
CGI-I (mean ± SD)	1.61 ± 0.97	2.80 ± 0.63
Etiology		
Medical condition, n (%)	21 (24%)	NA
Developmental history (ASD, ID), n (%)	26 (30%)	NA
Treatment		
Use of ECT, n (%)	11 (13%)	–
Efficacy of first line pharmacological treatment, n (%)	66 (75%)	–

Note: GAF: Global Assessment of Functioning scale; CGI-S: Clinical Global Impressions-Severity of Illness scale; CGI-I: Clinical Global Impressions-Improvement scale; ECT: Electroconvulsive therapy; catatonia was considered to be chronic if subjects had catatonic symptoms after discharge from the index episode. NA = Not available. Among non-catatonic subjects, other psychiatric diagnoses included: tics and related disorders (n = 5), obsessive compulsive disorder (n = 2), anxiety disorders (n = 3) attention deficit hyperactivity disorder (n = 4), disruptive disorders (n = 8).

using Student's t-test ($p < 0.05$). Fifth, as its discriminating threshold varied, the performance of the PCRS was measured by receiver operating characteristic (ROC) analysis. The data for the present study were analyzed using the statistical program R, version 2.12.2.

3. Results

3.1. Demographic and clinical characteristics

The socio-demographic, clinical and treatment characteristics of the subjects and controls are reported in Table 2. A total of 88 inpatients presented a diagnosis of catatonia, including 31 females (35%) and 57 males (65%). The mean age was 15.17 years. Most of the patients admitted would receive the diagnosis of catatonia-NOS since the underlying diagnosis was not immediately available at admission. At discharge, 55% of the participants have received the diagnosis of schizophrenia with catatonic feature and 38% the diagnosis of major mood disorder with catatonic features. In addition, catatonia occurred in 30% of the cases in subjects with autism or other developmental disorders. Twenty one of the 88 catatonic subjects (24%) presented a medical condition (auto immune condition: n = 5; infectious encephalitis: n = 1; epileptic encephalopathy: n = 3; iatrogenic encephalopathy: n = 2; genetic and metabolic condition: n = 10) (details are available in Consoli et al. (2012)).

3.2. Internal consistency

As presented Table 3 the most frequently reported catatonic sign was social withdrawal; in contrast, acrocyanosis occurred in only 10% of the sample. The PCRS sum scores ranged from 2 to 38 ($M = 21.87$, $SD = 7.5$) and the number of catatonic symptoms present in each subject ranged from 2 to 17 ($M = 10.7$, $SD = 2.79$). The score did not differ significantly between genders ($t(60) = 0.459$, $p = 0.695$) and was not correlated with age ($r = -0.026$, $p = 0.846$). Cronbach's alpha for total score was 0.67.

3.3. Construct validity

Total scores on the PCRS and GAF scales were moderately correlated ($r = -0.41$, $p = 0.001$) across the whole sample. However, the PCRS score did not predict the duration of stay ($r = 0.11$, $p = 0.402$). As expected, the PCRS and the ACE score were not correlated ($r = 0.03$, $p = 0.819$).

Table 3
Frequencies of each item and correlation with total PCRS score.

	PCRS scale				Present	rho
	0 "Absent"	1 "Occasional"	2 "Frequent"	3 "Constant"		
Catalepsy	30 (34%)	16 (18%)	26 (30%)	16 (18%)	66%	0.45[**]
Stupor	18 (20%)	12 (14%)	36 (41%)	22 (25%)	80%	0.46[**]
Posturing	24 (27%)	8 (9%)	35 (40%)	21 (24%)	73%	0.49[**]
Waxy flexibility	47 (53%)	15 (17%)	19 (22%)	7 (8%)	47%	0.41[**]
Staring	17 (19%)	17 (19%)	25 (28%)	29 (33%)	80%	0.53[**]
Negativism	10 (11%)	12 (14%)	22 (25%)	44 (50%)	89%	0.61[**]
Stereotypes	45 (51%)	15 (17%)	15 (17%)	13 (15%)	49%	0.25[*]
Psychomotor excitement	39 (44%)	18 (20%)	23 (26%)	8 (9%)	55%	0.00
Automatic compulsive movements	43 (49%)	13 (15%)	20 (23%)	12 (14%)	52%	0.10
Muscular rigidity	33 (38%)	19 (22%)	21 (24%)	15 (17%)	63%	0.57[**]
Social withdrawal	6 (7%)	5 (6%)	22 (25%)	55 (62%)	93%	0.56[**]
Mutism	22 (25%)	16 (18%)	19 (22%)	31 (35%)	75%	0.32[**]
Mannerism	47 (53%)	11 (12%)	14 (16%)	16 (18%)	46%	0.37[**]
Echopraxia	76 (86%)	4 (5%)	5 (6%)	3 (3%)	14%	0.37[**]
Echolalia	70 (80%)	3 (3%)	8 (9%)	7 (8%)	20%	0.30[**]
Incontinence	46 (52%)	10 (11%)	19 (22%)	13 (15%)	49%	0.39[**]
Verbigeration	57 (65%)	12 (14%)	15 (17%)	4 (5%)	26%	0.13
Schizophasia	74 (84%)	9 (10%)	3 (3%)	2 (2%)	15%	0.24[*]
Acrocyanosis	79 (90%)	3 (3%)	6 (7%)	0 (0%)	10%	0.19
Refusal to eat/drink	32 (36%)	16 (18%)	20 (23%)	20 (23%)	64%	0.43[**]

[**] $p < 0.001$.
[*] $p < 0.05$.

Table 4
Factor loading for the PCRS items.

Item (abbreviated)	Mean	SD	Factor 1	Factor 2	Factor 3	Factor 4
Staring	1.75	1.12	**0.59**	0.02	−0.13	−0.02
Social withdrawal	2.43	0.88	**0.57**	0.00	0.03	0.05
Negativism	2.14	1.04	**0.56**	0.14	0.25	−0.06
Muscular rigidity	1.20	1.13	**0.55**	0.24	0.10	−0.11
Refusal to eat/drink	1.32	1.19	**0.51**	0.18	0.01	−0.27
Mutism	1.67	1.20	**0.46**	−0.14	**−0.40**	0.03
Catalepsy	1.32	1.13	0.04	**0.87**	−0.05	0.11
Posturing	1.60	1.13	0.22	**0.52**	−0.06	0.12
Waxy flexibility	0.84	1.03	0.20	**0.52**	0.03	−0.07
Schizophasia	0.24	0.63	0.31	**−0.50**	−0.09	0.24
Stereotypes	0.95	1.13	0.10	0.00	**0.74**	0.12
Automatic compulsive movements	1.01	1.13	−0.04	−0.02	**0.72**	0.04
Mannerisms	0.99	1.20	0.28	−0.17	**0.46**	0.29
Stupor	1.70	1.06	0.39	0.19	**−0.49**	0.26
Echopraxia	0.26	0.72	−0.19	0.19	0.02	**0.72**
Echolalia	0.45	0.96	0.05	−0.05	0.10	**0.77**
Incontinence[*]	0.99	1.16				
Acrocyanosis	0.17	0.53	0.09	0.35	0.17	−0.19
Psychomotor excitement	1.00	1.04	−0.13	−0.36	0.20	0.29
Verbigeration	0.61	0.93	−0.32	0.14	0.08	0.31
Eigenvalues			2.38	2.29	1.96	1.72
Proportion of variance accounted for			13%	12%	10%	9%
Cumulative % of variance			13%	25%	35%	44%
Cronbach's alpha			0.73	0.71	0.70	0.61

Item loadings are bolded at a significance level of ≥0.40.

[*] Considering the very low degree of communality of the item "incontinence," it was omitted from further analysis.

3.4. Internal structure

An eigen value >1.0 and the screen test criteria indicated the relative suitability of either a four- or five-factor solution. The parallel analysis routine indicated that no more than four factors could be reliably extracted. The Velicer MAP criterion achieved a minimum of 0.03 with 4 factors. TLI was 0.944 and the RMSEA index was 0.054. Table 4 presents the results of an exploratory factor analysis with a weighted least squares orthogonal varimax solution. Four factors were extracted, which accounted for 44% of the variance. Factor 1 consisted of six catatonic symptoms associated with motor inhibition: staring, mutism, negativism, refusal to eat/drink, social withdrawal, and muscular rigidity. Factor 2 consisted of three signs associated with disturbances of volition: waxy flexibility, catalepsy, and posturing. As presented in Table 4, this factor was also correlated negatively with schizophasia. Factor 3 consisted of three abnormal involuntary movements: stereotypes, mannerisms, and automatic compulsive movements. This factor correlated negatively with stupor and mutism. Factor 4 consisted of echo phenomenon (or automatic imitation symptoms), including echolalia and echopraxia. Motor excitement, incontinence, verbigeration, and acrocyanosis did not load on a factor. The four factors showed modest correlations at best, with the highest between factors 1 and 4 ($r = 0.25$) and factors 1 and 3 ($r = −0.21$).

3.5. Catatonic factors across diagnostic group

The total PCRS score did not differ between the groups of patients. However, the schizophrenic group scored highest on factor 3 compared to the mood disorders group (Fig. 2a). Similarly, youths with developmental disorders scored higher in factor 3 items compared to those without associated developmental disorders (Fig. 2b). Other factors were found at a comparable rate across the diagnoses.

3.6. ROC analysis

Fig. 3 depicts the ROC curve corresponding to the way in which changes in the PCRS threshold influenced the classification of patients as exhibiting or not exhibiting catatonia from the total sample of 138 patients. The discriminant validity of the PCRS showed a higher cut-off score of 9 (AUC = 0.983, sensitivity = 0.97, specificity = 1). The discriminant validity of the scale was similar when the control group only contained the subjects treated with antipsychotic drugs (AUC = 0.978, sensitivity = 0.95, specificity = 1).

4. Discussion

4.1. Validity of the PCRS and internal structure

The internal consistency was moderate. As expected, the PCRS score correlated with the measure of global functioning, and no significant association was found with adverse life events. Analysis of the internal structure of the PCRS showed that pediatric catatonic syndrome is composed of several distinct dimensions. Previous studies conducted in adults have identified two (Abrams et al., 1979; Morrison, 1973), three (Wilson et al., 2015), four (Kruger et al., 2003; McKenna et al., 1991; Northoff et al., 1999; Ungvari et al., 2007) or six (Peralta and Cuesta, 2001) dimensions of catatonic symptoms. Such a discrepancy can arise from methodological differences between studies due to the diversity in samples (e.g., including or not including somatic diseases), the nature of the scale used (Sienaert et al., 2011), and the method of statistical extraction (Muthén and Kaplan, 1985). The current findings should therefore be first interpreted with regard to studies that used the BFCRS scale (Ungvari et al., 2007; Wilson et al., 2015).

Ungvari et al. (2007) examined the structure of the BFCRS among 225 inpatient adults (mean age 41.7) with chronic schizophrenic disorders. A four-factor model explained 50% of the variance, and the authors distinguished between a negative/withdrawal factor (16%), an automatic phenomena factor (12%), a repetitive/echo phenomena factor (12%), and an agitated/resistive factor (10%). Wilson et al. (2015) presented results from principal component analysis of the BFCRS among 339 inpatients (mean age 38.4). A three-factor model best fit the data and accounted for 37% of the total variance. The identified factors were increased (16%), abnormal (14%), and decreased (7%) psychomotor activity.

The first factor identified in our sample encompassed symptoms marked by motor inhibition (such as mutism, negativism, and social withdrawal). It is roughly comparable with the "negative/withdrawal" factor isolated by Ungvari et al. (2007) and the "decreased psychomotor activity" factor isolated by Wilson et al. (2015). The analyses of the internal structure of other scales support the predominance of this dimension in catatonic syndrome. The first "negativism/stupor" factor, which accounted for 32% of total variance of an eight-item catatonic checklist, was isolated (Abrams et al., 1979), along with the first "motor poverty" factor, which accounted for 23% of the variance of the Modified Rogers Scale (MRS) (Peralta and Cuesta, 2001). This dimension could be predominant among subjects with retarded catatonia (Morrison, 1973). An independent dimension of motor inhibition symptoms in youths with catatonic episode is also supported by clinical evidence. Indeed, youths with pervasive refusal syndrome show a pattern of acute catatonic symptoms that belongs almost exclusively to this dimension (e.g., mutism, negativism, and refusal to eat) without other types of catatonic symptoms (Dhossche and Kellner, 2015).

The second factor is composed of symptoms of catalepsy (i.e., maintenance of posture). This is somewhat analogous to the BFCRS factors of "automatic" motor activity (Ungvari et al., 2007) and "abnormal psychomotor activity" (Wilson et al., 2015). The analyses of the internal structure of the Catatonia Rating Scale (CRS) (Kruger et al., 2003) and the Northoff Catatonia Scale (NCS) (Northoff et al., 1999) showed that these symptoms form an independent dimension,
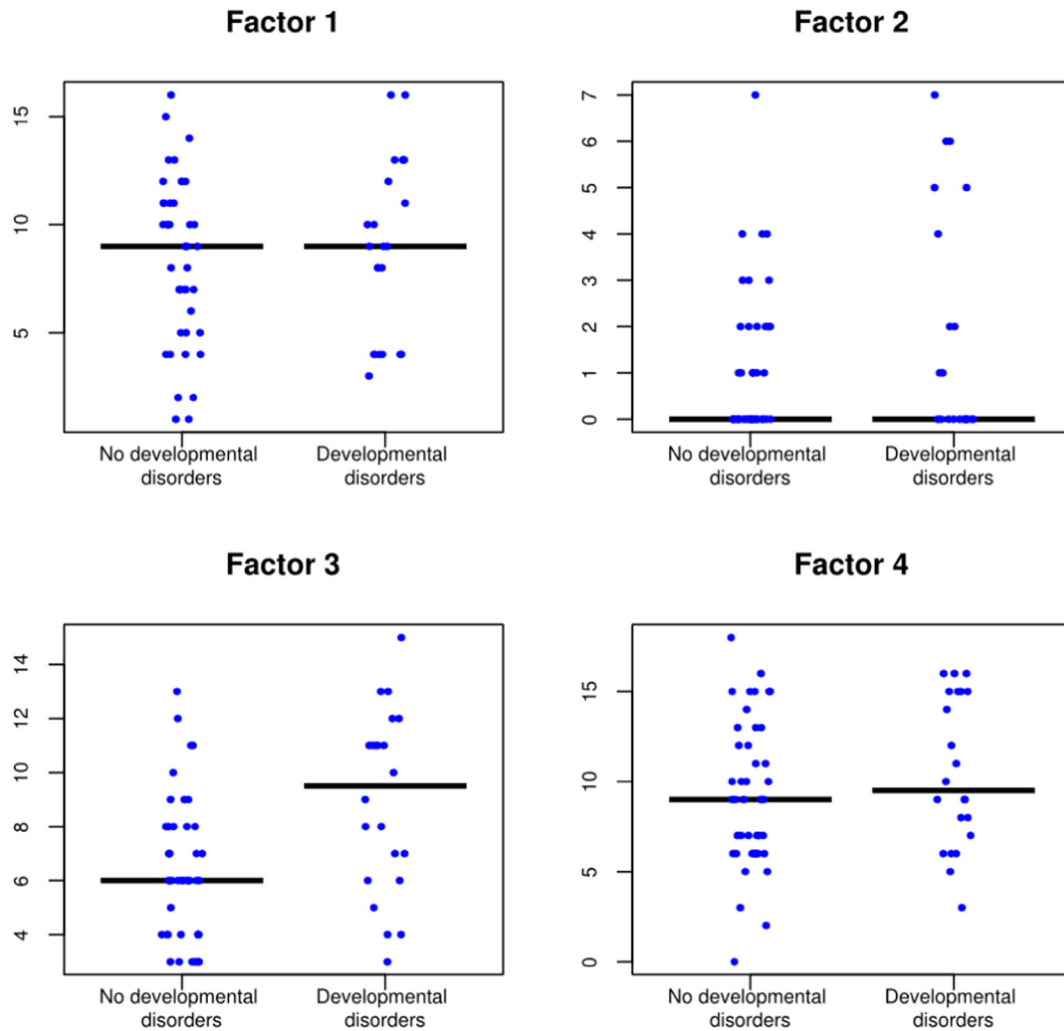
Fig. 2. a. Comparison of the means of each of the four factors between youths with and without developmental disorders. b. Comparison of the means of each of the four factors between youths with mood disorders and those with schizophrenic disorders.
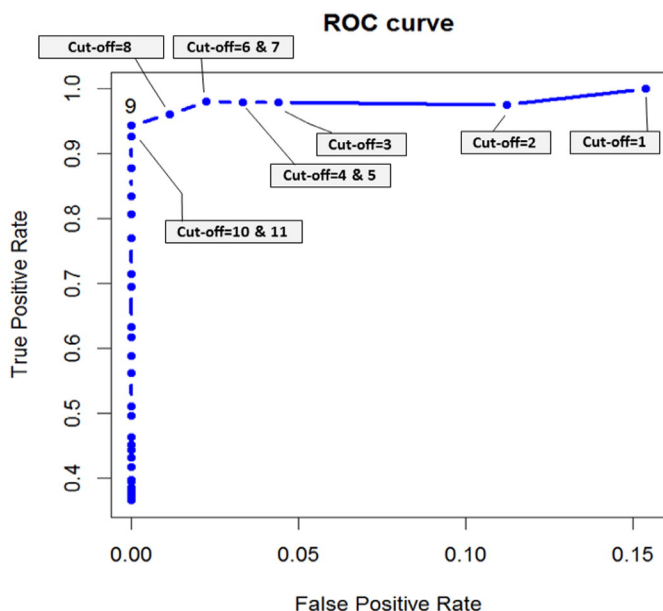

Fig. 3. ROC analysis.

distinct from catatonic symptoms associated with negativism and withdrawal; however, the study conducted by Peralta and Cuesta (2001) contradicted this finding.

The third dimension identified was composed of bizarre and repetitive movements. This is partially comparable with the BFCRS dimension of "repetitive/echo" catatonic symptoms (Ungvari et al., 2007) and more alike the "abnormal involuntary movements/mannerisms" factor (Kruger et al., 2003), the third "stereotypes/mannerisms" factor of the MRS (Peralta and Cuesta, 2001) and the second "hyperactive/excited" factor of the NCS (Northoff et al., 1999).

The fourth group of symptoms was composed of echo phenomena. Some authors, such as Ungvari et al. (2007) and Wilson et al. (2015), consider the echo phenomena to be distinct from other dimensions, whereas others thought that it should be included in a wider "abnormal motor activities" dimension (Kruger et al., 2003; Northoff et al., 1999; Peralta and Cuesta, 2001).

All 4 factors were remarkably univocal; that is, they did not share symptoms with other factors to a significant degree. Compared to previous studies in adults, we did not find that specific factors reflected hyperactive symptoms or excitement. This could be explained by the relatively low prevalence of mania in this sample (Cohen et al., 1999; Cornic et al., 2009) as reported in the literature (Takaoka and Takata, 2003).

4.2. Catatonic factors across diagnostic groups

The schizophrenic patient group and those with a history of developmental disorders scored higher on factor 3 compared to other patient groups. In line with this, Kruger et al. (2003) found that adults with schizophrenia were more likely to present abnormal involuntary movements and mannerisms when experiencing catatonia compared to those diagnosed with mood disorder. However, in the present study, the way that the distribution of catatonic symptoms varied across diagnoses was not pathognomonic, as factor 3 was also rather high in other diagnoses. The fact that factors in general had weak diagnostic specificity shows that catatonia should be considered as a syndrome and supports recent changes in the DSM-5 criteria (American Psychiatric Association, 2013; Cohen et al., 1999).

4.3. ROC analysis

The PCRS scored low in a group of inpatients with psychiatric disorders without catatonic syndrome. The use of the scale in a control group is relevant to ensure that aberrant motor functioning symptoms due to a developmental disorder or substance-induced are not systematically rated as catatonic symptoms. However, in order to test the validity of the PCRS in screening catatonia among inpatient youths, subjects and controls should have been selected following a similar process. Indeed, we cannot determined here if the differences observed between catatonic and control subjects with respect to socio-demographic and clinical characteristics result from the clinical specificities of pediatric catatonia, the characteristics of the psychiatric disorder associated, or selection bias due to distinct selection process. The finding from ROC analysis should therefore be regarded as preliminary and, importantly, the threshold at 9 cannot be generalized to other samples without further investigations.

4.4. Strengths and limitations

The results of this study should be interpreted in the context of its limitations. First, the number of subjects recruited was low, despite the 22-year recruitment period; this can be due to the relatively low prevalence of catatonia in youths (Cohen et al., 2005; Thakur et al., 2003). Therefore, the low subjects-to-item ratio may have affected the quality of the factorial analysis. Second, these data did not allow for the assessment of inter-rater and test-retest reliability. Third, the relation between specific catatonic dimensions and psychiatric disorders could reflect symptom overlap, as the clinical assessment was cross-sectional. We used the psychiatric diagnosis retained at discharge while catatonic symptoms were collected at admission to limit the potential bias. Fourth, our study presents sources of heterogeneity that may not be generalizable. In particular, our clinical sample of acutely ill patients recruited in a university teaching hospital can be particularly enriched in subjects with more severe forms of catatonia. As shown for the BFCRS in adults (Wilson et al., 2015), the reliability of the PCRS might substantially vary according to the severity of catatonic symptoms. Future research should examine whether the psychometric features of the PCRS are comparable in youths with milder forms of catatonia, such as brief toxic-related catatonic episodes reported in emergency unit (Masi et al., 2002; Maxwell et al., 1993). However, unlike previous studies (except (Wilson et al., 2015), our data included both somatic- and psychiatric-related catatonic episodes and then covered diverse aspects of the syndrome. Fifth, as mentioned above, finding from the sensitivity analysis is difficult to interpret. Basically, ROC analysis was performed to make sure that the PCRS does not score excessively in non-catatonic youths with or without antipsychotics.

4.5. Clinical and research implications

Most importantly, the notion that catatonic symptoms do not differ considerably between diagnoses in youths has to be kept in mind for clinical evaluation in psychiatric settings. Next studies could help determine whether the presence of certain catatonic symptoms is useful for prognostic assessment. One can also ask whether a better knowledge of catatonic dimensions and their clinical correlates might guide therapeutic interventions. As almost one-quarter of catatonic inpatient adolescents do not respond to a second line of treatment (Raffin et al., 2015), it would be helpful to identify these subjects on the basis of their symptoms. Thus, posturing predicted a lower response to benzodiazepine in adults (Ungvari et al., 1999) and adolescents (Raffin et al., 2015) with catatonic syndrome. Finally, repeated assessment of symptoms before and throughout pharmacological treatment could highlight the mechanisms of motor disturbances that underpin the dimensions of catatonic symptoms.

5. Conclusion

In this study, we tested the psychometric characteristics of a pediatric catatonic scale without preliminary assumptions about underlying psychiatric or somatic diagnoses. The internal and external validity of the PCRS was acceptable. Factor analyses showed a four-factor solution in line with previous findings in adults: a "negative withdrawal" factor, a "catalepsy" factor, an "abnormal movements" factor and an "echo phenomenon" factor. However, unlike adults, no "hyperactive/excitement" dimension of catatonic symptoms was identified. The structure of catatonia symptoms was found to be roughly comparable across the different groups (i.e., affective disorders, schizophrenia, and developmental disorders), confirming that catatonia should be regarded as a syndrome in young patients as well.

Appendix A. The Pediatric Catatonic Rating Scale

1. Catalepsy:	11. Withdrawal:
Passive induction of a posture held against gravity	Refusal to make eye contact and not responding to nonverbal communication
0 = Absent	0 = Absent
1 = Less than 1 minute	1 = Occasional
2 = Greater than one minute, less than 15 minutes	2 = Frequent
3 = More than 15 minutes	3 = Constant
2. Stupor:	12. Mutism:
Extreme hypoactivity, immobile, minimally responsive to stimuli	No, or very little, verbal response, not applicable if there is an established aphasia
0 = Absent	0 = Absent
1 = Sits abnormally still, may interact briefly	1 = Verbally unresponsive to majority of questions, incomprehensible whispers
2 = Virtually no interaction with external world	2 = Speaks less than 20 words/5 min
3 = Non-reactive to painful stimuli	3 = No speech

3. Posturing:

Active and/or spontaneous maintenance of a posture against gravity
0 = Absent
1 = Less than 1 minute
2 = Greater than one minute, less than 15 minutes
3 = More than 15 minutes

4. Waxy flexibility:

Slight and even resistance to positioning by examiner
0 = Absent
3 = Present

5. Staring:

Fixed gaze, little or visual scanning of environment, decreased blinking
0 = Absent
1 = Poor eye contact, decreased blinking
2 = Gaze held longer than 20 s, occasionally shifts attention
3 = Fixed gaze, non-reactive

6. Negativism:

Opposing or not responding to instructions or external stimuli
0 = Absent
1 = Mild resistance and/or occasionally contrary
2 = Moderate resistance and/or frequently contrary
3 = Severe resistance and/or continually contrary

7. Stereotypes:

Repetitive, abnormally frequent, non-goal directed movements
0 = Absent
1 = Occasional
2 = Frequent
3 = Constant

8. Excitement:

Extreme hyperactivity, constant motor unrest which is apparently non purposeful. Not to be attributed to akathisia or goal directed agitation
0 = Absent
1 = Excessive motion
2 = Constant motion, hyperkinetic without rest periods
3 = Full-blown catatonic excitements, endless frenzied motor activity

9. Automatic compulsive movements:

Involuntary muscle activity exhibited in posture, attitudes, mimic or gesture due to inhibition or forced motor action
0 = Absent
1 = Occasional
2 = Frequent
3 = Constant

10. Rigidity:

Maintenance of a rigid position despite efforts to be moved, exclude if cogwheeling or tremor present
0 = Absent
1 = Mild resistance
2 = Moderate
3 = Severe, cannot be repostured

13. Mannerisms:

Odd caricature of normal actions
0 = Absent
1 = Occasional
2 = Frequent
3 = Constant

14. Echopraxia:

Mimicking another's movements
0 = Absent
1 = Occasional
2 = Frequent
3 = Constant

15. Echolalia:

Mimicking another's speech
0 = Absent
1 = Occasional
2 = Frequent
3 = Constant

16. Incontinence:

Nocturnal enuresis, daytime urinary incontinence, or fecal incontinence
0 = Absent (or no recent worsening)
1 = Nocturnal and/or occasionally diurnal
2 = Frequently diurnal
3 = Constant

17. Verbigeration:

Repetition of phrases or sentences, like a scratched record
0 = Absent
1 = Occasional
2 = Frequent
3 = Constant

18. Schizophasia:

Scrambled speech, word salad, seemingly random words and phrases linked
0 = Absent
1 = Occasional
2 = Frequent
3 = Constant

19. Acrocyanosis:

Cyanosis of the extremities
0 = Absent
1 = Occasional
2 = Frequent
3 = Constant

20. Refusal to eat, drink:

Severe decrease of daily food or drink intake
0 = Absent
1 = Minimal per os intake for less than 1 day
2 = Minimal per os intake for more than 1 day
3 = No per os intake for one day or more

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