ORIGINAL ARTICLE

Defining the optimal threshold scores for catatonia spectrum (CS) in clinical and general population

Liliana Dell'Osso[1], Giulia Amatori[1], Ivan Mirko Cremone[1], Enrico Massimetti[2], Benedetta Nardi[1], Barbara Carpita[1]

[1] Department of Clinical and Experimental Medicine, University of Pisa, Pisa, Italy; [2] UFSMA Val di Cornia, Azienda USL Toscana Nord-Ovest

Summary

Background. The catatonia spectrum (CS) questionnaire is a recently developed tool designed to evaluate a broad spectrum of catatonic manifestations, including both full-threshold and sub-threshold symptoms. While the CS has proven useful for quantifying catatonic symptoms, it currently lacks a validated diagnostic threshold.

Aim. This study aimed to determine the optimal cut-off score on the CS for identifying the presence of clinically significant catatonia.

Methods. The study included 182 individuals diagnosed with catatonia (CTN) and healthy controls (HC). Trained psychiatrists conducted clinical diagnoses based on Diagnostic and Statistical Manual of Mental Disorders (DSM-5) criteria and subsequently assessed participants using the CS. The Propensity Score procedure was used with one-to-one correspondence to balance the diagnostic groups compared on the basis of gender and age, resulting in a sample composed of 79 subjects with catatonia and 79 healthy controls. A receiver-operating characteristics (ROC) curve was used to find the optimal CS threshold score for the identification of full-blown catatonia.

Results. A cut-off score of 30 on the CS proved most effective in distinguishing individuals with catatonia, exhibiting satisfactory levels of both specificity and sensitivity.

Conclusion. Our findings suggest that a CS score of 30 serves as the most discriminative threshold for identifying individuals with catatonia.

Keywords: catatonia, catatonia spectrum, catatonia questionnaire, CS cut-off score

INTRODUCTION

The first description of catatonia dates back to 1874, when the German psychiatrist Karl Ludwig Kahlbaum used this term to define a distinct, cyclical, and progressive illness characterized by alternation between stupor and excitement [1], along with behavioral and motor manifestations (negativism, mutism, stereotypes, mannerisms, automatic obedience, automatism, impulsivity, and agitation) combined with cognitive, affective, and neurovegetative symptoms. This definition was challenged and partly reformulated by Emil Kraepelin (1856-1926), who in his seminal work "Compendium der Psychiatrie" first published in 1883 [2], distinguished two forms of psychosis: manic depression and dementia praecox, considering catatonia as one of the possible forms of the latter. Kraepelin's conceptualization of mental disorders exerted an unprecedented influence on the course of psychiatry, becoming the basis for what was reported in the first edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM) [3]. A similar fate befell catatonia, as it remained relegated for about a century to the realm of psychoses. Within the fifth edition of DSM (DSM-5) and its text revision (DSM-5-TR) [4], catatonia has been placed in the "Schizophrenia Spectrum and Other

Psychotic Disorders chapter", alongside schizophrenia, other psychotic disorders, and schizotypal personality disorder. It is a complex neuropsychiatric syndrome characterized by a constellation of psychomotor signs and symptoms developed in the context of numerous pathological conditions, not only of psychiatric nature but also neurological, toxic, endocrinological, and infectious [5]. The diagnosis is met by the presence of three or more criteria among the following: stupor (absence of motor activity or no active relation with the environment); catalepsy (passive induction of a posture maintained against gravity); Waxy flexibility (slight constant resistance to a postural modification induced by the examiner); Mutism (absent or very scarce verbal response); negativism (oppositivity or no response to external stimuli or instructions); posturing (spontaneous and active maintenance of a posture against gravity); mannerism (artificial and extravagant caricatures of normal actions); stereotypy (repetitive purposeless movements, abnormally frequent); agitation not influenced by external stimuli; grimacing (strange facial expressions not appropriate to the context); echolalia; echopraxia [5]. Catatonic symptoms are nonspecific and can be found in other mental disorders. The DSM-5 does not conceive catatonia as an autonomous category but rather as a transnosographic specifier, describing three distinct conditions: "catatonia associated with another mental disorder": a neurodevelopmental disorder, a psychotic disorder, bipolar disorder, depressive disorder, or another mental disorder; "Catatonic disorder due to a medical condition": neurological (neoplasms, head trauma, cerebrovascular disease, encephalitis), metabolic (hypercalcemia, hepatic encephalopathy, homocystinuria, diabetic ketoacidosis), and others; "Unspecified catatonia": a provisional diagnosis to be used when the nature of the underlying disorder is unclear or there is insufficient information (for example, in an emergency room setting) or when the criteria for catatonia are not fully met [5]. This third category, including conditions not fully meeting the criteria necessary for diagnosis according to DSM-5, involves the identification of a "subthreshold catatonia," opening the doors to defining a "catatonia spectrum". In this scenario, a self-assessment questionnaire named catatonia spectrum (CS) was created to investigate the symptoms associated with the catatonic spectrum through the use of 74 items. Validated in 2022 [6], the CS demonstrated a excellent internal consistency and test-retest reliability, along with strong convergent validity when compared to other dimensional measures of catatonia. The aim of the present study is to determine the optimal cut-off score on the CS for identifying the presence of clinically significant catatonia.

MATERIALS AND METHODS

Data have been collected between November 2021 and January 2022 at the Department of Psychiatry of the Azienda Ospedaliero Universitaria Pisana (AOUP).

Study sample and procedures

The present study involved 182 subjects endorsing at least 3 symptom criteria for catatonia (CTN) and individuals without current or lifetime mental disorders (healthy controls, HC) and belonging to health care and paramedical personnel, all evaluated according to DSM-5 diagnostic criteria. Exclusion criteria were: age under 18 years, language or intellectual impairment affecting the possibility to fulfill the assessments, mental disability, poor cooperation skills, and ongoing psychotic symptoms. All subjects were aged 18-60 years old and signed a written informed consent. The Structured Clinical Interview for DSM-5, Research Version (SCID-5-RV) [7] was used to confirm the diagnoses of catatonia, as well as the absence of mental disorders among healthy controls. The study was conducted in accordance with the Declaration of Helsinki. The Ethics Committee of the AOUP approved all recruitment and assessment procedures (Date of approval: 27/10/2021; ID number: 20608). Eligible subjects provided written informed consent, after receiving a complete description of the study and having the opportunity to ask questions. Subjects were not paid for their participation according to Italian legislation.

Measures

The CS questionnaire is a self-assessment tool designed to explore various manifestations of the catatonic spectrum, including core, subthreshold, atypical, and partial symptoms experienced throughout life. It is structured into eight domains, each addressing different aspects such as psychomotor activity, verbal response, repetitive movements, artificial expressions, oppositivity to stimuli, response to external instructions, automatisms, and impulsivity. With a total of 74 items, respondents provide dichotomous answers ("Yes" or "No") to each question. In the validation study [6], the CS questionnaire demonstrated exceptional internal consistency and test-retest reliability, along with robust convergent validity when compared to other dimensional measures of catatonia, such as the Bush-Francis Catatonia Rating Scale (BFCRS) and the Bush-Francis Catatonia Screening Instrument (BFCSI) [8].

Statystical analyses

We performed Student t-tests and Chi-square tests in order to compare mean age and sex among groups, respectively.

To balance the diagnostic groups compared on the basis of gender and age, the Propensity Score procedure was used with one-to-one correspondence. This procedure matches the records of the case group (in our case, the catatonic group) with similar records of the control group. It first performs a logistic regression with the diagnostic group variable as the dependent variable and gender and age as independent variables. Then select a match for each case in the control group based on the propensity score from logistic regression. The score is an estimate of the probability of belonging to the case group. For the score in the controls and in the corresponding cases, we specified a tolerance of 0.5. The procedure produces and activates a new dataset of matching cases and controls. A receiver-operating characteristics (ROC) curve was utilized to find the threshold value of CS total score that best identify the subjects meeting the DSM-5 criteria for full-blown catato-

nia. According to the definition of accuracy, the ROC analysis was built on the basis of sensitivity and specificity for different cut-offs. Statistical analyses were conducted using SPSS version 26.0 [9].

RESULTS

We recruited a total of 182 subjects. At the end of balancing procedures, the sample resulted in 158 individuals (Females 63.9%, N = 101; Males 36.1%, N = 57; mean age: 40.99±11.99), distributed in two groups: 79 patients with catatonia (Females: 62.0%, N = 49; Males: 38.0%, N = 30; mean age: 42.05 ± 13.132) and 79 healthy controls (HC group; Females: 65.8%, N = 52; Males: 34.2%, N = 27; mean age: 39.94 ± 10.462). The two diagnostic groups did not significantly differ from each other in terms of age (p = 0.683; F = 0.167) and sex (p = 0.619, X^2 = 0.247) (Tab. I).

According to the ROC analysis, the most discriminant cutoff of the CS total score for identifying subject with catatonia was 30, with an AUC value of 0.817 (p < .001), a sensitivity of 0.72, a specificity of 1.00 (Fig. 1) and a Youden Index of 0.722 (Fig. 1).

DISCUSSION

The objective of this research was to determine a threshold score on the catatonia screening (CS) questionnaire for iden-
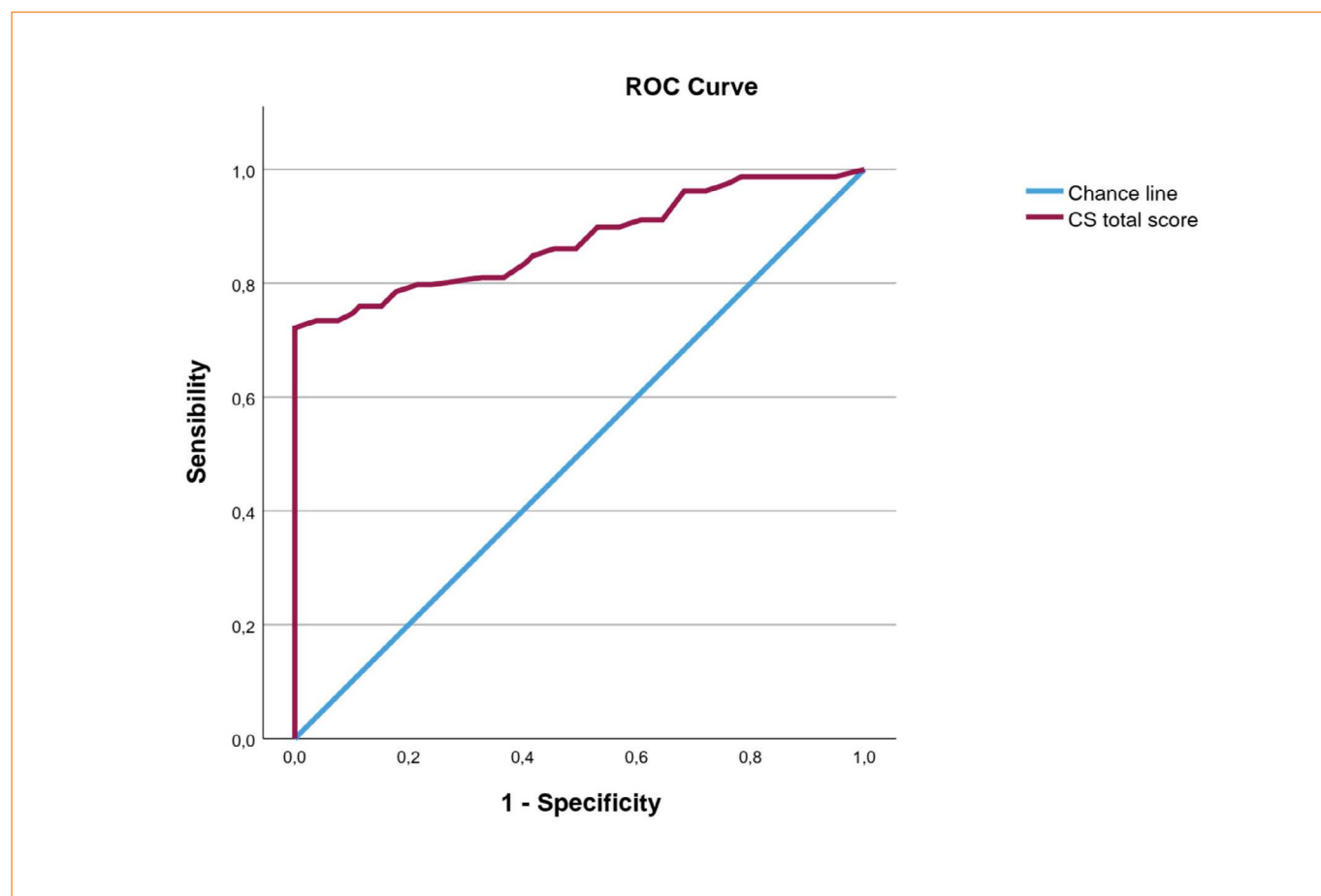

FIGURE 1. ROC curve of the CS score for determining catatonia. AUC = 0.817; Youden Index = 0.722; threshold score = 30.

TABLE I. Comparison of sociodemographic variables amongst groups.

		Catatonia (N = 79)	Healthy Controls (N = 79)		
		Mean ± DS	Mean ± DS	F	p
Age		42.05 ± 13.132	39.94 ± 10.462	0.167	0.683
		N(%)	N(%)	*Chi-Square*	*p*
Sex	*F*	49(62.0)	52(65.8)	0.247	0.619
	M	30(38.0)	27(39.9)		

tifying catatonia in adults. Our findings suggest that a score of 30 on the CS, indicating positive responses to 30 questionnaire items, is the most effective threshold. This score demonstrated good levels of specificity (0.72) and sensitivity (1.00). In clinical practice, using the CS could enhance prevention and diagnosis of catatonia, a potentially life-threatening condition, across various patient populations with mental and medical disorders. Furthermore, the use of the CS questionnaire has proven particularly useful thus far in studying the relationships between catatonia, autism, and mood disorders [10,11], providing intriguing insights into the understanding of such psychopathological conditions Hence, the availability of a threshold value is poised to expand the range of feasible research in this domain, while also serving as a valuable asset in everyday clinical settings. However, the study has limitations. It involved a relatively small sample size with limited demographic characteristics considered. Additionally, while it identified a threshold for distinguishing the presence or absence of catatonia symptoms, it did not address distinguishing between significant subthreshold catatonic traits. Hence, the CS may not fully assess subthreshold catatonic symptoms. Moreover, since the CS is self-reported, it may be prone to bias. Therefore, it should not be solely relied upon for diagnosis in clinical settings. Nonetheless, based on these findings, the CS can serve as a useful screening tool to confirm and assist in clinical diagnosis among individuals suspected of catatonia.

CONCLUSION

The best threshold value for the identification of catatonia trough the CS questionnaire was a score of 30. This value was associated with good levels of specificity (0.72) and sensibility (1.00).

References

1. Kahlbaum K. Die Katatonie Oder das Spannungsirresein. Berlin, Germany: Hirschwald; 1874.

2. Kraepelin E. Psychiatrie, V Auflage, Ein lehrbuch fur studierende und artze. Leipzig: Barth 1896.

3. Mayes, R, Horwitz AV. DSM-III and the revolution in the classification of mental illness. Journal of the history of the behavioral sciences 2005;41(3):249-267. https://doi.org/10.1002/jhbs.2010

4. American Psychiatric Association: Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition. Arlington, VA: American Psychiatric Association 2013.

5. American Psychiatric Association: Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition Text Rev. Arlington, VA: American Psychiatric Association 2022.

6. Dell'Osso L, Amatori G, Cappelli A, et al. Catatonia Spectrum: Validation of a Questionnaire Investigating Catatonia Spectrum 2022:13:913286. https://doi.org/10.3389/fpsyt.2022.913286

7. First MB, Williams JBW, Karg RS, et al. Structured Clinical Interview for DSM-5-Research Version (SCID-5 for DSM-5, Research Version; SCID-5-RV). Arlington, VA: American Psychiatric Association 2015.

8. Bush G, Fink M, Petrides G, et al. Catatonia. I. Rating scale and standardized examination. Acta Psychiatr Scand 1996;93(2):129-136. https://doi.org/10.1111/j.1600-0447.1996.tb09814.x.

9. IBM Corp. IBM SPSS Statistics for Windows, Version 26.0. Armonk, NY: IBM Corp; 2019.

10. Dell'Osso L, Amatori G, Massimetti G, et al. Investigating the relationship between autistic traits and symptoms and Catatonia Spectrum. Eur Psychiatry 2022;65(1):e81. https://doi.org/10.1192/j.eurpsy.2022.2334

11. Dell'Osso L, Amatori G, Carpita B, et al. The mediating effect of mood spectrum on the relationship between autistic traits and catatonia spectrum. Front Psychiatry 2023;14:1092193 https://doi.org/10.3389/fpsyt.2023.1092193