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The Population-Based Incidence and Prevalence of Catatonia

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Abstract

Objectives: Catatonia is a neuropsychiatric disorder that is associated with a range of medical and psychiatric illnesses. While many single-center studies have been conducted, there remains uncertainty in the population-based incidence and prevalence of the disorder. This study reports the incidence and prevalence of catatonia extrapolated from two large epidemiologic studies in the United Kingdom (UK) and United States (US).

Methods: Incidence rates (defined as the number of catatonic episodes beginning per 100,000 person-years) and prevalence rates (defined as the proportion of individuals with catatonia in a given year) are calculated.

Results: Based on UK data, there was an incidence of 4.34 (95% CI 3.98 to 4.72) catatonic episodes per 100,000 person-years with an average 1-year prevalence of 4.39 (95% CI 4.03 – 4.77) per 100,000 persons. US data demonstrates a 1-year prevalence of 5.15 (95% CI: 5.08 – 5.23) catatonia hospitalizations per 100,000 persons.

Conclusions: Catatonia is a rare disorder, qualifying as an orphan disease under both European Medicines Agency and US Food and Drug Administration criteria. Further research is needed to rigorously define the epidemiology of catatonia in other populations.

Catatonia is a neuropsychiatric disorder characterized by psychomotor, speech, and affective pathology. Understanding of the disorder has evolved since its first description in the 1870s, as has its place in psychiatric classification (1). Present criteria using the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition, Text Revision (DSM-5-TR) and the International Classification of Diseases 11th Revision (ICD-11) identify catatonia as occurring with a range of other medical and psychiatric disorders as well as without any associated condition (2). These evolving diagnostic criteria have contributed to challenges in defining the prevalence of the disorder, particularly as there may be trade-offs in the sensitivity and specificity of catatonia diagnosis depending on the number and type of diagnostic criteria required. A 2018 meta-analysis examined data from 110,774 participants across 80 independent studies from 1935 to 2017 to estimate a pooled catatonia prevalence of 9.0% among clinical samples (3). Notably, the smaller included studies reported significantly higher prevalences than larger studies, the meta-analysis was unable to provide estimates of catatonia incidence and prevalence in the population overall. The authors concluded that "large studies of representative samples are needed in order to further inform the prevalence range and moderator variables of catatonia" (3). Since the publication of this meta-analysis, two large population-based studies have been published using unrelated methods in two countries. The first by Rogers *et al*. used an analysis of the full text of medical records from a region in London, United Kingdom (UK) (4), while the second by Luccarelli *et al*. used administrative claims records from a national database of hospitalizations in the United States (US) (5). In order to better characterize the epidemiology of catatonia, this manuscript estimates the population-based incidence and prevalence of catatonia-associated hospitalizations and medical visits, extrapolating from these data sources.

Methods

The incidence and prevalence of catatonia are derived from Rogers *et al*. using episodes of catatonia occurring in any care setting (including inpatient psychiatry and medical-surgical hospitals) meeting *DSM-5-TR* criteria, to reflect present case definitions. Incidence is defined as new cases of *DSM-5-TR* catatonia occurring during the study period. As catatonia generally resolves between episodes, for the purposes of incidence calculations, each episode is treated separately—that is, a patient with two distinct episodes of catatonia would contribute twice to incidence calculations. This is akin to how incident rates of many infections disease are calculated, where recurrent infections in the same individual are counted equally to single infections in separate individuals (6). To calculate incidence, the number of episodes of catatonia that started during a 10-year follow-up period (from 2007 to 2016, inclusive) was divided by the total person-years in the regional population.

Period prevalence is defined as the proportion of a population that exhibit catatonia at any time over a given period; this includes newly incident cases as well as those who persist in a catatonic state but were diagnosed earlier. This 1-year prevalence is thus defined as the number of unique individuals experiencing catatonia within a calendar year divided by the regional population in that year. This prevalence is presented as the average of the 1-year prevalence rates over the 10-year study period.

In a complementary approach, the prevalence of catatonia hospitalizations was extrapolated from inpatient hospitalizations in the US as reported by Luccarelli *et al*. For this dataset, catatonia was defined based on *International Statistical Classification of Diseases, Tenth Revision, Clinical Modification* (*ICD-10-CM*) discharge diagnosis codes F06.1 or F20.2. This study was conducted during the era of DSM-5 diagnoses, but the precise criteria used to diagnose catatonia in each hospitalization are unknown. The prevalence rates derived by Luccarelli *et al*.

from hospitalizations in a dataset including 5,139 hospitals and 32,355,827 hospitalizations were extrapolated to include all hospitalizations in the American Hospital Association (AHA) 2020 survey (33,356,853 admissions among 6,093 hospitals) (7) and the US Census population in 2020 (331,449,281) based on the 2020 US census (8). Recurrent hospitalizations in the year for the same individual are indistinguishable in this dataset, and so an individual may be counted more than once in this prevalence calculation.

Additional methodological details are given in Table S1. Incidence and prevalence figures were calculated with 95% confidence intervals using the binomial exact method, with calculations performed using Stata/MP (version 15.1). The CRIS system has approval from the Oxfordshire C Research Ethics Committee (ref: 18/SC/0372) with data analysis approved by the CRIS Oversight Committee (ref: 17-102). Analysis of the NIS has been declared Not Human Subjects Research by the Mass General Brigham IRB.

Results

Rogers *et al*. identified 539 catatonic episodes among 373 individuals among 12,420,547 person-years of follow-up. This represents an incidence of 4.34 (95% CI 3.98 to 4.72) catatonic episodes per 100,000 person-years. During the same period there were 545 total catatonic episodes, for an average 1-year prevalence of 4.39 (95% CI 4.03 – 4.77) per 100,000 persons (Table S2). Luccarelli *et al*. identified 16,575 catatonia hospitalizations in the US in 2020, with an overall rate of 0.0512% of hospitalizations involving catatonia. Extrapolating this to the total number of US hospitalizations based on 2020 AHA data represents 17,079 catatonia hospitalizations in a population of 331,449,281, for a prevalence of 5.15 (95% CI: 5.08 – 5.23) catatonia hospitalizations per 100,000 person-years.

Discussion

The incidence and prevalence of catatonia were strikingly similar in two large and independent datasets describing catatonia epidemiology, with an estimated 1-year prevalence rate of 4.39 (95% CI 4.03 – 4.77) per 100,000 person based on UK data and 5.15 (95% CI: 5.08 – 5.23) hospitalizations per 100,000 person-years based on US data. As an individual may be counted more than once in the US data, this estimate represents an upper bound on prevalence, but the true rate is expected to be lower.

The similar incidence and prevalence rates for catatonia reflects an overall short duration of catatonic illness. While the duration of catatonic episodes has not been rigorously studied, studies have identified chronic catatonia lasting >5 years (9), but one large-scale study in India found a mean duration of episodes of 84 days (10), and duration of illness of approximately 6 months was sufficiently long to merit case report (11), suggesting that mean duration of illness is shorter than this, which is consistent with the results reported here.

Diseases are defined as orphan by the European Medicines Agency if they have a prevalence of less than 50 per 100,000, and by the US Food and Drug Administration if they affect fewer than 200,000 Americans (a prevalence of 60.3 per 100,000 in 2020), meaning that catatonia meets orphan disease criteria by a 10-fold margin for both the US and European Union. Even assuming substantial under-diagnosis of catatonia (with one prospective study from an inpatient psychiatric unit finding that 63.8% of catatonia cases were not identified using DSM-5 criteria) (12), this would still classify catatonia as an orphan disease under US and EU definitions.

The epidemiologic results reported here are difficult to compare to prior meta-analytic results derived from smaller studies generally examining psychiatric inpatients (3). Applying the meta-analytic 9.0% rate of catatonia to 2020 survey results of the Substance Abuse and Mental Health Services Administration in the US, which identified 77,622 psychiatric inpatients in the US on one day in 2020 (13), would imply 6,986 psychiatric inpatients with catatonia on that day (2.11 per 100,000 Americans) which is potentially consistent with the numbers reported here, but this would require further focused study. Furthermore, additional research is needed to develop optimal operationalized definitions of catatonia, including the impact of diagnostic criteria (including symptom clusters and motor vs. affective symptoms) on rates of catatonia diagnosis, for instance comparing case rates using ICD-11 vs DSM-5 criteria (2).

Strengths of this study include the inclusion of two large population-based studies relying on different methodology (administrative claims and chart review) from health systems in two countries. Reassuringly, the overall incidence and prevalence numbers derived from both sources are similar, despite the differing methodologies and treatment settings. Limitations include the retrospective design of these studies, meaning that any misdiagnosis (either underdiagnosis or overdiagnosis) would bias estimates, and the rate of this cannot be assessed from these data sources. In particular, for the UK data of Rogers *et al.* if treating clinicians did not assess for catatonia then these features could not be identified in the full text of clinical notes, and for the US data of Luccarelli *et al.* the administrative claims data in unable to determine what criteria were used to diagnose catatonia. Finally, the population incidence and prevalence estimates here do not account for potential demographic differences in diagnosis by age, race, or comorbidities, and thus may not apply directly to nations beyond the US and UK.

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