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Original Research Article

The State of the Catatonia Literature: Employing Bibliometric Analysis of Articles From 1965–2020 to Identify Current Research Gaps

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Background: *Since Kahlbaum's classic 19th-century description of catatonia, our conceptualization of this syndrome, as well treatment options for it, has advanced considerably. However, little is known about the current state of the catatonia literature since a comprehensive bibliometric analysis of it has not yet been undertaken.* **Objective:** *The purpose of this study was to conduct a bibliometric analysis, along with a content analysis of articles reporting new findings, to better understand the catatonia literature and how catatonia research is changing.* **Methods:** *Using the search term "Title(catatoni*)" in Web of Science Core Collection for all available years (1965–2020), all available publications (articles, proceeding papers, reviews) pertaining directly to catatonia were identified, and metadata extracted. Semantic and coauthorship network analyses were conducted. A content analysis was also conducted on all available case reports, case series, and research articles written in English.* **Results:** *A total of 1015 articles were identified representing 2861 authors, 346 journals, and 15,639 references. The average number of publications per year over the last 20 years (31.3) more than doubled in comparison to that in the 20 years prior (12.8). The top 3 most common journals were* Psychosomatics/ Journal of the Academy of Consultation-Liaison Psychiatry*,* Journal of ECT*, and* Schizophrenia Research*, which represented 12.6% of all publications. Content analysis revealed that catatonia articles are increasingly published in nonpsychiatric journals. There was a notable paucity of clinical trials throughout the study period. Since 2003, articles on catatonia secondary to a general medical condition, as well as articles including child/adolescent patients and patients with autism spectrum disorder or intellectual disability, have made up increasing shares of the literature, with a smaller proportion of articles reporting periodic or recurrent catatonia. We noted a decrease in the proportion of articles detailing animal/*in vitro *studies, genetic/heredity studies, and clinical trials, along with stagnation in the proportion of neuroimaging studies.* **Conclusions:** *The catatonia literature is growing through contributions from authors and institutions across multiple countries. However, recent growth has largely been driven by increased case reports, with significant downturns observed in both clinical and basic science research articles. A dearth of clinical trials evaluating potential treatments remain a critical gap in the catatonia literature.*

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Bibliometric Analysis of Catatonia

INTRODUCTION

Psychiatry has made considerable gains in better understanding the pathophysiology of catatonia and treating this complex neuropsychiatric behavioral syndrome in the nearly 150 years since its nosological birth. However, despite these advances, catatonia continues to frequently evade the detection of psychiatrists and other clinicians in child/adolescent[1] and adult patient populations in both psychiatric[2] and general medical settings,[3] hindering delivery of appropriate care and often leaving patients with catatonia without a name for the collection of anomalous behaviors they have been experiencing. Recently, an international study evaluating the ability of psychiatrists, psychiatry trainees, and medical students revealed that participants correctly answered only 55% of test questions on catatonia and identified only 69% of Bush-Francis Catatonia Rating Scale (BFCRS) items on a standardized patient exam.[4] Somewhat disconcertingly, despite presumably more exposure to catatonic patients and training in its diagnosis, psychiatrists only slightly outperformed medical students on both evaluations.

With this in mind, it is apparent that patients with catatonia stand to benefit considerably from the medicine field working to advance its understanding of this syndrome and improve clinical training on catatonia diagnosis and treatment. Fundamental to our efforts to meet these goals is the need for a more comprehensive understanding of the existing catatonia literature, including how the literature is changing, where important gaps continue to lie, and to what degree catatonia has penetrated nonpsychiatric academic journals due to ever-increasing recognition of catatonia associated with general medical conditions. Such an understanding might improve efforts to build upon previous findings with new insights and data by highlighting facets of catatonia that have been underinvestigated, as well as infrequently employed methodologies in catatonia research.

Evaluating the academic literature in particular areas for trends has now become much easier via bibliometrics, the quantitative statistical analysis of documents. Bibliometrics is increasingly being employed as a tool to examine scientific publications and collect information about their authors, journals of publication, cited sources, and impact. Readers have likely already encountered commonly used bibliometric output measures such as article citation count or author "h-index." While useful for more clearly visualizing the often-opaque landscapes of the academic literature, bibliometrics' reliance on these flawed and manipulable impact metrics mean that it often does not capture important contributions to a field by scholars who may produce a low number of units of scholarly output, making it no substitute for careful qualitative evaluation.[5] However, bibliometrics can quickly facilitate the creation of a broad overview of the literature in a particular area, as well as which people and institutions are heavily influencing it, providing a unique vantage point from which to view not only the long-term evolution of a literature but also areas for expansion.

In recent decades, bibliometrics has increasingly been employed to investigate research on a variety of mental health conditions and their treatment, including addiction,[6] bipolar disorder,[7] depression,[8] eating disorders,[9] personality disorders,[10] obsessive compulsive disorder,[11] schizophrenia,[12] and suicide.[13] Additional studies have investigated other areas of contemporary interest in psychiatry such as psychedelics,[14,15] machine learning,[16] lithium toxicity,[17] ketamine,[18] and neuroimaging.[19] However, no bibliometric analyses have yet been published on catatonia. To help address this gap in the bibliometric literature, we performed a bibliometric analysis of the catatonia literature from 1965 through 2020. Here we present our findings on the catatonia literature, including where it is being published, who is generating it, and how it is changing, while also highlighting areas in need of more research.

MATERIALS AND METHODS

Search Strategy and Analysis

In a search conducted on June 21, 2021, the search term "Title(catatoni*)" was used to identify documents related to catatonia in the Web of Science Core Collection for all available years (1965–2020). A search based on title, rather than keyword or other criteria, was chosen to better ensure that the articles identified primarily, and specifically, pertained to catatonia. The use of the wildcard* term additionally allowed for the identification of articles using other common terms within this body of literature (e.g., catatonia and catatonic). Similar search term strategies have been employed in other systematic reviews on the catatonia literature.[20] We included all published articles and reviews in our search, but all other publication types aside

from journal articles (e.g., books, conference abstracts, etc.) were excluded.

R-statistical software (version 4.0.5) was used to complete the bibliometric analysis with the open-source R-tool *bibliometrix*[21] and its associated Web-based app *biblioshiny*. This software was used to complete the descriptive analysis of this body of work and was subsequently employed to evaluate articles, citations per article, authors, articles per author, articles per country, corresponding author institution and country, journals of publication, and references within articles. The most cited papers, authors, institutions, and countries were identified. We also identified the most cited references within the data set for each of 4 roughly 50-year-long time periods (1818–1869, 1970–1919, 1920–1869, and 1970–2020).

Network Analyses

We conducted network analyses to study the pattern of coauthor collaboration and semantic relationships in publications. For coauthor collaboration, we constructed a network where nodes were defined as individual authors, and edges (links) were defined as the presence/number of shared publications (or coauthorships) between authors. Next, author affiliation country was extracted from publication author lists and matched. Nodal degree was calculated as the sum of a node's connections (weighted) and subsequently summed per country. For the semantic analysis, we constructed a network where nodes were defined as individual terms present in the abstract section of publications, and edges were defined as the number of co-occurrence network of terms (e.g., 2 terms that frequently occur together in an abstract would have a stronger connection in the network). We discarded irrelevant terms (e.g., "initial", "included", "mg/kg") and merged synonymous/equivalent terms (e.g., EEG and electroencephalography). We then used a network community detection algorithm to identify clusters or "communities" of nodes. The algorithm groups nodes that are highly interconnected together and less connected to the rest of the network.[22]

We used Gephi 0.9.2 (https://github.com/gephi/gephi) and Cytoscape 3.9.1 (https://github.com/cytoscape) for visualization.[23,24]

Content Analysis

All accessible articles written in English and reporting new findings (case reports, case series, and research study) underwent a content analysis by author B. S. B. to determine the following characteristics for each: article type (case report, case series, and research study); type of journal published in psychiatric/psychological/neuroscience; substance use disorder; general medical (including substance use disorder) and nonmedical conditions associated with reported cases of catatonia (psychiatric conditions, other medical conditions [including substance use], and substance use); inclusion of co-occurring catatonia and delirium case; inclusion of malignant catatonia, neuroleptic-induced catatonia, or neuroleptic malignant syndrome (NMS) case; inclusion of recurrent, relapsing, or periodic catatonia case; inclusion of child/adolescent patients; inclusion of geriatric (65 years of age or older) patients; inclusion of patients with autism or intellectual disability; inclusion of peripartum patients; reports findings from animal/*in vitro* study; reports findings from genetic/heredity study; reports findings from interventional clinical trial; reports findings on catatonia education/evaluation of clinician knowledge; reports neuroimaging findings; reports treatments besides lorazepam and electroconvulsive therapy (ECT); reports treatment with ECT; or discusses inflammation hypothesis for catatonia.

To assess temporal differences in these characteristics, articles were divided into 2 cohorts by median year of publication (2003), with the "old cohort" containing articles published from 1965 to 2002 and the "new cohort" containing articles published from 2003 to 2020. Pearson's chi-squared test was performed using Stata version 15.1 (StataCorp LLC, College Station, TX). Statistical significance was set at $P < 0.05$. Since this analysis was exploratory, there was no correction for multiple comparisons.

RESULTS

Retrieved Articles, Citations, and Publication Rate

Our analysis produced 1015 articles written by 2861 authors, published in 346 journals, and cited a total of 18,200 times. The mean time ± standard deviation since publication was 18.4 ± 14.39 years prior to our search. A breakdown of the number of articles according to year is illustrated in Supplemental Figure 1. The mean number of citations per article was 17.85 ± 14.71, and mean annual citations per document was 1.03 ± 0.52. The 10 most cited articles are presented in Table 1. With

Bibliometric Analysis of Catatonia

TABLE 1. The 10 Most-Cited Articles on Catatonia

Rank	Title	Authors	Journal	Year	Total citations	Conclusion	Publication type
1	Catatonia. I. Rating scale and standardized examination.[25]	Bush G, Fink M, Petrides G, Dowling F, Francis A.	*Acta Psychiatrica Scandinavica*	1996	448	The Bush-Francis Catatonia Rating Scale, a 23-item rating scale, demonstrates good inter-rater reliability and construct validity. The first 14 items can be used to screen for catatonia.	Clinical study
2	The catatonic syndrome.[26]	Gelenberg A.J.	*Lancet*	1976	291	Catatonia, which is often assumed to be a subtype of schizophrenia, is rather a syndrome with various possible causes.	Survey of history and literature.
3	Catatonia in psychiatric classification: a home of its own.[27]	Taylor M.A. and Fink M.	*American Journal of Psychiatry*	2003	282	Catatonia can be distinguished from other behavioral syndromes by a cluster of clinical features and should be considered as an individual category in psychiatric diagnostic systems.	Literature review
4	Catatonic syndrome in a general psychiatric inpatient population: frequency, clinical presentation, and response to lorazepam.[28]	Rosebush P.I., Hildebrand A.M., Furlong B.G., Mazurek M.F.	*Journal of Clinical Psychiatry*	1990	258	Lorazepam administered to patient with signs of catatonia showed dramatic clinical improvements.	Prospective clinical study
5	Catatonia. A prospective clinical study.[29]	Abrams R. and Taylor M.A.	*Archives Of General Psychiatry*	1976	258	Catatonic signs are nonspecific, homogeneously distributed among a variety of clinical diagnostic entities, and do not predict treatment response.	Prospective clinical study
6	Lethal catatonia.[30]	Mann S.C., Caroff S.N., Bleier H.R., Welz W.K., Kling M.A., Hayashida M.	*American Journal of Psychiatry*	1986	235	Lethal catatonia is a syndrome, rather than a specific disease, and represents an uncommon, but deadly, neuropsychiatric condition.	Literature review
7	Catatonia. II. Treatment with lorazepam and electroconvulsive therapy.[31]	Bush G., Fink M., Petrides G., Dowling F., Francis A.	*Acta Psychiatrica Scandinavica*	1996	234	Lorazepam was an effective primary treatment for catatonia with ECT as an effective salvage treatment option.	Prospective clinical study
8	Catatonic reactions to high-potency neuroleptic drugs.[32]	Gelenberg A.J. and Mandel M.R.	*Archives Of General Psychiatry*	1977	195	8 Patients receiving high-potency neuroleptic drugs developed an acute syndrome of catatonia and parkinsonism	Case report

TABLE 1. (*Continued*)

Rank	Title	Authors	Journal	Year	Total citations	Conclusion	Publication type
9	Catatonia in autistic spectrum disorders.[33]	Wing L. and Shah A.	*The British Journal of Psychiatry*	2000	178	Catatonia is a later complication seen in autistic spectrum disorders (6% total, 17% in patients older than 15 y).	Cross-sectional survey
10	What catatonia can tell us about "top-down modulation": a neuropsychiatric hypothesis.[34]	Northoff G.	*Behavioral and Brain Sciences*	2002	161	Comparing Parkinson disease and catatonia uncovers importance of possible bi-directional modulation between cortical and subcortical structures.	Case reports, literature review

ECT = electroconvulsive therapy.

448 citations, the most cited article was the one by Bush and colleagues in 1996 introducing the BFCRS.[25] The growth rate in the publication of articles on catatonia for the whole period was 580% (with an average annual growth rate of 32.1%), with articles from 2000 to 2020 constituting 65.28% of publications identified (with an average annual growth rate of 7.64% for that 20-year period).

Academic Institutions, Authors, Countries, and Journals

The mean number of authors per publication was 2.82, with only 123 (12.22%) articles being single-authored. The 5 corresponding author countries with the most total number of articles published were the United States (286), Germany (74), France (53), Japan (43), UK (39), and China (34). The 5 journals publishing the most catatonia articles were *Psychosomatics (Journal of the Academy of Consultation-Liaison Psychiatry)* (N = 55, 5.43%), *Journal of ECT* (N = 49, 4.84%), *Schizophrenia Research* (N = 24, 2.37%), *Journal of Clinical Psychiatry* (N = 23, 2.27%), and *Acta Psychiatrica Scandinavica* (N = 19, 1.88%). Supplemental Table 1 details the 10 journals publishing the most catatonia articles. Figure 1 illustrates how rates of publication on catatonia have changed in the journals with the largest number of catatonia publications during the study period.

Cited Sources

The 1015 identified articles cited 15,639 total references. The earliest referenced source was Heinroth's 1818 textbook.[35] To capture influence throughout different years of the literature, we identified the most-cited references published within each of 4 roughly 50-year-long time periods (1818–1869, 1870–1919, 1920–1969, and 1970–2020) and have included this in Supplemental Table 2. The most-cited article of the 19th-century was Karl Kahlbaum's initial description of catatonia,[36] and that of the 20th-century was the article by Bush and colleagues describing the BFCRS.[25]

Network Analysis

The coauthorship network was limited to 1000 nodes. There was a mixture of authors that only collaborate locally, as well as groups that span multiple countries (Figure 2A). The countries with the largest number of authors (nodes) were the United States (US), Germany, France, Japan, Italy, and Russia (Figure 2B). Countries with the largest number of connections (per country sum of nodal degree) were Germany, US, France, Italy, and Japan (Figure 2B). Countries with the largest number of authors were not necessarily the ones with the largest number of connections (e.g., US versus Germany; Japan versus Italy). The median number of coauthorships was 7. Most authors had between 5 and 10 coauthorships (Figure 2C). High-degree nodes (hubs)—i.e., authors with the highest number of coauthorships—made up a very small portion of all authors (Figure 2D).

After processing, the semantic network consisted of 325 nodes. The community detection algorithm identified 11 modules (represented using different colors in

FIGURE 1. Stratification of Catatonia Articles by Journal Published From 1965 to 2020

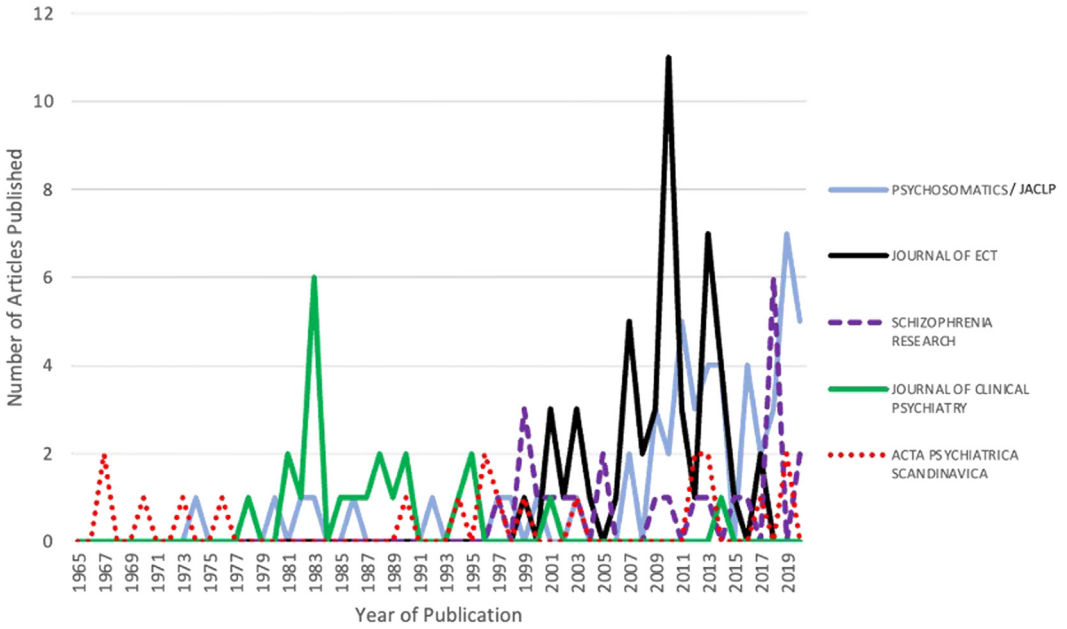


Figure 3A). A number of these modules follow a thematic distribution. For example, there are discernible modules that are treatment-related (teal), neurological/medical (brown), NMS-related (red), core symptoms/signs (blue), etc. (Figure 3A). High-degree nodes (hub-like terms) are featured in Figure 3B and include psychiatric conditions most associated with catatonia (mood/affective disorders and schizophrenia), common presenting features (mutism, negativism, psychomotor changes), NMS, and the most common treatments (ECT and lorazepam). Similar to the coauthorship network, high-degree nodes made up a very small portion of all nodes (Figure 3C).

Content Analysis

Of the 1015 articles in the bibliometric analysis data set, 906 were manually reviewed to determine whether they reported new findings and could be included in a subsequent content analysis (109 articles were excluded from this review due to being written in a language other than English [104] or being inaccessible [5]). The breakdown of article type among these 906 articles was 50.1% (454) case report, 26.8% (243) research article, 16.2% (147) review, 6.7% (61) case series, and 2.2% (20) other (total percent sums to greater than 100 due to some reviews and research articles also containing case reports within them). Of the total, 720 (79.5%) articles were published in psychiatric, psychological, behavioral science, or neuroscience journals, 183 (20.2%) in other types of medical journals, and 0.3% (3) in nonmedical journals.

To assess how characteristics of articles on catatonia reporting new findings are changing, further analysis was restricted to case reports, case series, and research articles (n = 755). The median year of publication for this corpus was 2003. Therefore, we analyzed how article characteristics proportionally differed between an old cohort (published from 1965 to 2002) and a new cohort (published from 2003 to 2020). This analysis revealed, among other observations, that the proportion of catatonia articles published in psychiatric, psychological, behavioral science, or neuroscience journals fell from 84.9% in the old cohort to 75.9% in the new cohort (P = 0.002), while the proportions published in other types of medical journals (inclusive of substance use disorder journals) rose from 15.1% to 22.7% (P = 0.007). The proportion published in substance use disorder journals rose from 0% to 2.2% (P = 0.003). Paralleling this trend, the proportion of articles reporting cases of catatonia associated with another mental disorder fell from 62.8% in the old cohort to 54.8% in the new cohort (P = 0.025), while the proportion of articles

FIGURE 2. **Coauthorship Network. *A*: General landscape of coauthorship network, nodes represent authors, edges represent links (the presence/number of coauthorship), and colors represent country of affiliation. Smaller author subnetworks typically comprise single countries (localized to the peripheries in [*A*]), while larger networks comprise cross-country collaborations (closer to the center in [*A*]; enlarged in [*D*]). *B*: Number of authors and total number of connections (degree) by country. Note that countries with the largest number of authors are not necessarily the ones with the largest number of connections (e.g., US versus Germany). *C*: Node degree distribution. Most authors have between 5 and 10 coauthorships. Note that high-degree nodes (i.e., "hubs") make up a very small portion of all authors. *D*: Three representative subnetworks that illustrate different patterns of collaboration (taken from [*A*]). The subnetwork on the left represents a 2-country collaboration (US-Germany), the subnetwork in the middle has the largest diversity of affiliation countries, and the one on the right represents a case where a central hub node is an author with a different affiliation country.**

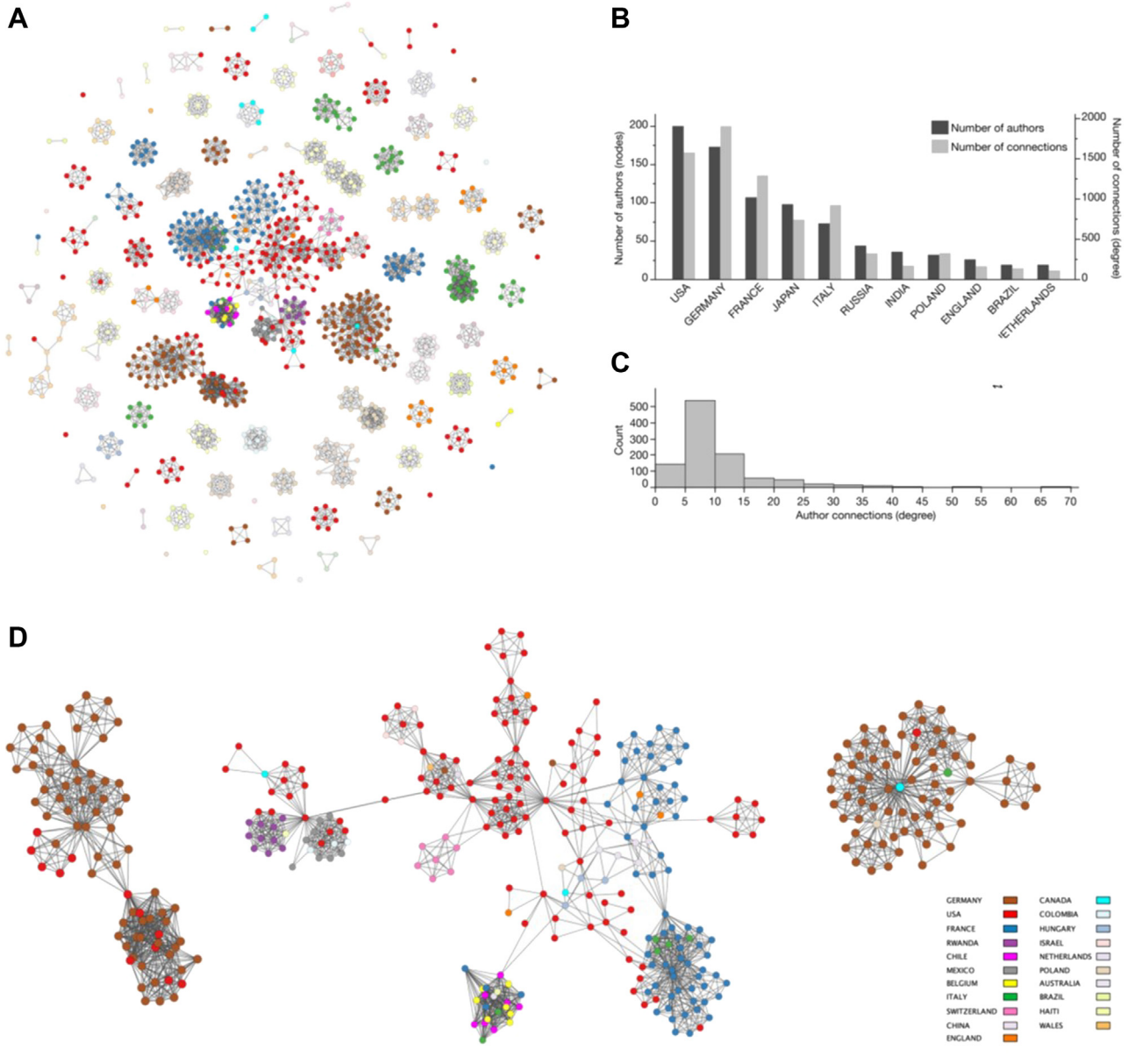
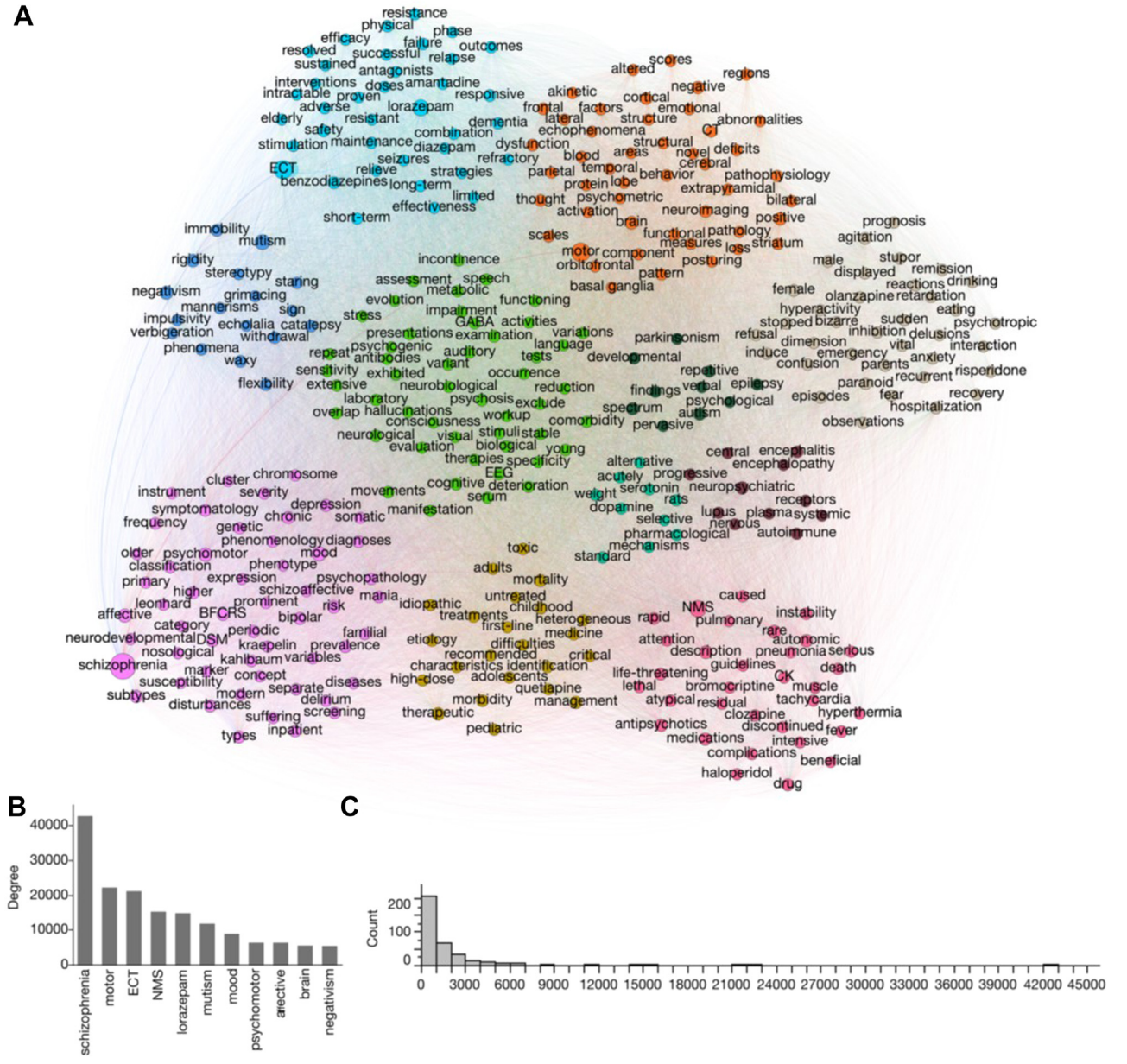


FIGURE 3. **Semantic Network. *A*: Semantic co-occurrence network, nodes represent different terms, edges represent terms co-occur (width is proportional to co-occurrence frequency). Nodal diameter is proportionate to the nodal degree (sum of edges). The network was parsed using a community detection algorithm that resulted in 11 modules represented using different colors. Note the thematic distribution—e.g., treatment-related (*teal*), neurological/medical (*brown*), NMS-related (*red*), core symptoms/signs (*blue*). *B*: Nodes with the highest degree which represent the most central terms. *C*: Node degree distribution. ECT = electroconvulsive therapy; NMS = neuroleptic malignant syndrome.**



reporting catatonic disorder associated with another medical condition (inclusive of substance use) rose from 36.7% to 51.2% ($P < 0.001$). The proportion of articles reporting catatonia associated with substance use ranged from 2.6% to 5.8% ($P = 0.027$).

Case reports constituted a larger share of the new cohort compared to the old cohort (67.4% versus 53.3%, $P < 0.001$). While there was no statistically significant change for case series (7.4% old cohort versus 8.8% new cohort, $P = 0.502$), the proportion of research studies

significantly dropped (39.7% old cohort versus 24.1% new cohort [$P < 0.001$]). Increases were noted in the proportion of articles in the new cohort examining child/adolescent patients (17.0% versus 11.3%, $P = 0.024$) and patients with autism or intellectual disability (12.1% versus 6.4%, $P = 0.007$), while the proportion reporting cases of periodic, recurrent, or relapsing catatonia fell (8.5% versus 18.7%, $P < 0.001$). There was no change in the proportion reporting cases of malignant catatonia, neuroleptic induced catatonia, or NMS (8.8% versus 11.3%, $P = 0.251$). There were increases in the new cohort of papers with a focus on catatonia education and assessing clinician knowledge about catatonia (1.1% versus 0%, $P = 0.038$), while there were decreases in the proportion of articles reporting findings from animal/*in vitro* studies (3.3% versus 9.5%, $P = 0.001$), genetic/heredity studies (1.6% versus 4.9%, $P = 0.013$), and interventional clinical trials (0.6% versus 2.1%, $P = 0.071$). There was no change in the proportion of articles reporting neuroimaging findings (5.5% new cohort versus 5.1% old cohort, $P = 0.83$), and this was also true when looking at only neuroimaging findings reported as part of a research study (2.2% new cohort versus 2.1% old cohort, $P = 0.893$). Further details on these and other article characteristics are summarized in Table 2.

Of the 10 articles reporting prospective interventional clinical trials, 3 included randomized, placebo-controlled trials.[37–39] Of note, the study by Marténi and colleagues reports pooled results from open-label and double-blind clinical trials, leaving only 2 articles reporting findings exclusively from randomized, placebo-controlled trials of treatments for catatonia.[39] We are aware of 1 additional randomized, placebo-controlled trial examining the efficacy of ECT versus risperidone for nonaffective catatonia resistant to lorazepam that was not identified by our search strategy, bringing the total number of randomized, placebo-controlled trials for catatonia treatments to 4.[40]

DISCUSSION

With 1015 articles published from 1965 to 2020, the catatonia literature appears smaller or likely to be smaller than the literature on antisocial personality disorder (8154 articles from 1980 to 2019),[10] bipolar disorder (4270 articles from 1980 to 2004),[41] borderline personality disorder (8901 articles from 1980 to 2019),[10] delirium (3591 articles from 1995 to 2015),[42] depression (76,293 articles from 2004 to 2019),[43] eating disorders (37,446 articles from 198 to 2020),[9] postpartum depression (717 articles from 2000 to 2020),[8] schizophrenia (67,893 articles from 1950 to 2006),[44] and schizotypal personality disorder (2706 articles from 1980 to 2019)[10] but possibly larger than the literature on avoidant, dependent, histrionic, narcissistic, obsessive compulsive, paranoid, and schizoid personality disorders (less than 400 articles on each published from 1980 to 2019).[10] While the relative size of the catatonia literature is as expected when compared to more common psychiatric conditions such as depression, with an estimated prevalence of 8%–38% in psychiatric patients,[45] it appears that catatonia has received a relatively small amount of attention within the psychiatric literature. To put this in perspective further, with 1241 articles published from 1913 to 2016 (nearly all from the late 1940s to 2016), it appears the literature on lithium toxicity alone may be larger than the catatonia literature.[17] Of note, our search strategy did not include terms associated with catatonia, such as *katatonia*, akinetic mutism, *délire aigu*, delirious mania, Bell's acute exhaustive mania, acute maniacal delirium, and NMS, that may have expanded the catatonia corpus somewhat.

Importantly, the number of academic publications on catatonia annually produced appears to have dramatically increased in recent decades, with over 60% of catatonia publications being published in the last 20 years. Previously published analyses examining the growth rates of the scientific literature at large suggests growth rates approximating 8%–9% for most of the 20th century.[46] This analysis indicates that the growth of articles on catatonia has far exceeded the typical expected growth over the entire analysis period (average annual growth rate of 32.1%) but has slowed down and practically mirror these expected rates over the last 20 years (average annual growth rate of 7.64%). The number of articles on catatonia published per year in the last 20 years is much higher than that in the past but appears to have stabilized from past periods of faster growth. Some of this rise in overall number of publications is likely accounted for by a nonspecific increase in avenues for academic publications (particularly open-source academic journals that are more willing to publish case reports) as well as possibilities concerning limited digitization of the scientific literature from past decades. Reasons for a recent stabilization, or possible slowing, in growth remain more elusive.

TABLE 2. Content Analysis of Case Reports, Case Series, and Research Studies Comparing Old (1965–2002) and New (2003–2020) Cohorts

Article characteristic	Entire sample (n = 755)	Old cohort (n = 390)	New cohort (n = 365)	P value
Article type: case report*	454 (60.1)	208 (53.3)	246 (67.4)	**<0.001**
Article type: case series*	61 (8.1)	29 (7.4)	32 (8.8)	0.502
Article type: research article*	243 (32.2)	155 (39.7)	88 (24.1)	**<0.001**
Journal type: psychiatric/psychological/neuroscience	608 (80.5)	331 (84.9)	277 (75.9)	**0.002**
Journal type: other medical (including substance use disorder)	147 (19.5)	59 (15.1)	88 (24.1)	**0.002**
Journal type: substance use disorder	8 (1.1)	0 (0)	8 (2.2)	**0.003**
Catatonia associated with another† mental disorder	445 (58.9)	245 (62.8)	200 (54.8)	**0.025**
Catatonic disorder due to another† medical condition	330 (43.7)	143 (36.7)	187 (51.2)	**<0.001**
Includes catatonia associated with substance use	31 (4.1)	10 (2.6)	21 (5.8)	**0.027**
Includes co-occurring catatonia and delirium	20 (2.7)	8 (2.1)	12 (3.3)	0.290
Includes neuroleptic induced catatonia, malignant catatonia, or NMS	76 (10.1)	44 (11.3)	32 (8.8)	0.251
Includes periodic, recurrent, or relapsing catatonia	104 (13.8)	73 (18.7)	31 (8.5)	**<0.001**
Includes child/adolescent patients	106 (14.0)	44 (11.3)	62 (17.0)	**0.024**
Includes geriatric patients	54 (7.2)	29 (7.4)	25 (6.9)	0.755
Includes patients with autism/intellectual disability	69 (9.1)	25 (6.4)	44 (12.1)	**0.007**
Includes peripartum patients	13 (1.7)	7 (1.8)	6 (1.6)	0.873
Reports findings from animal or *in vitro* study	49 (6.5)	37 (9.5)	12 (3.3)	**0.001**
Reports findings from genetic/heredity study	25 (3.3)	19 (4.9)	6 (1.6)	**0.013**
Reports findings from interventional clinic trial	10 (1.3)	8 (2.1)	2 (0.6)	**0.071**
Reports findings on catatonia education/evaluation of clinician knowledge	4 (0.5)	0 (0)	4 (1.1)	**0.038**
Reports neuroimaging findings	40 (5.3)	20 (5.1)	20 (5.5)	0.830
Reports neuroimaging findings (research study only)	16 (2.1)	8 (2.1)	8 (2.2)	0.893
Reports treatment other than lorazepam or ECT	214 (28.3)	115 (29.5)	99 (27.1)	0.471
Reports treatment with ECT	189 (25.0)	87 (22.3)	102 (28.0)	0.074
Discusses inflammation hypothesis of catatonia	4 (0.5)	0 (0)	4 (1.1)	**0.038**

Bold font in "P value" column indicates statistically significance at $P < 0.05$ level.
ECT = electroconvulsive therapy; NMS = neuroleptic malignant syndrome.
* Sums to greater than 100% due to some research study article also including case reports.
† Sums to greater than 100% due to some articles reporting catatonia of both types.

Notably, *Psychosomatics* (*Journal of the Academy of Consultation-Liaison Psychiatry* as of 2021) has become the most popular journal for articles on catatonia in the last decade, while there has been a significant drop in annual catatonia publications within the *Journal of ECT* (Figure 2), possibly a result of

prominent catatonia researcher Max Fink no longer serving as editor of that journal. Notably, we found that the absolute number of publications on catatonia increased from 2003–2020 compared to 1965–2002 although this was not statistically significantly.

Our coauthorship network analysis revealed that while there is strong collaboration between groups in some countries such as the US and Germany, the field of catatonia research may benefit from expansion of international collaborations. On semantic network analysis, we found the highest degree node was centered on the term "schizophrenia", a likely reflection of the now dispelled, although still influential, notion that catatonia is only encountered as a subtype of schizophrenia.

Our content analysis revealed a number of intriguing findings about the state of the catatonia, some causes for concern, and others causes for hope. The growth of articles reporting catatonia associated with other medical conditions, including substance use, may be a sign of increasing diagnosis and treatment of catatonia in nonpsychiatric populations, supported by increased proportions of catatonia articles published in nonpsychiatric medical journals and substance use disorder journals in the new cohort of catatonia articles (2003–2020). Growing recognition of autoimmune encephalitis, frequently associated with catatonia, might also be contributing to the increase in articles on catatonia associated with a general medical condition.[47] Recently published updated treatment guidelines for the treatment of catatonia in the medically ill patients[48] may help improve publication rates on catatonia in this once-overlooked population. One area of particular progress on this front is recent systematic investigation of catatonia occurring in patients with delirium.[49] However, while we observed a small increase in the proportion of articles in the new cohort reporting cases of co-occurring catatonia and delirium, it was not a statistically significant difference. Given the high rate of co-occurring catatonia and delirium, it is clear that more studies are needed to improve detection and treatment of catatonia in delirious patients, as well as a reconsideration of DSM-5 criteria preventing diagnosis of catatonic disorder due to another medical condition occurring exclusively during the course of a delirium.[49,50]

Increasing proportions of articles including child/adolescent patients and patients with autism spectrum disorder or intellectual disability in the new cohort suggest increasing recognition of catatonia in important populations where it had previously been overlooked. This is particularly important given the difficulties in communicating their distressing experiences that some members of these populations may have even prior to developing the communicative difficulties catatonia often brings. An increase in the proportion of articles focused on catatonia education and assessing clinicians' knowledge of catatonia also offers further hope, in that there appears to be increased focus on improving clinicians' ability to diagnose and treat catatonia.

There was a nonstatistically significant fall in articles detailing catatonia in geriatric populations. Given the aging of the world population, particularly in industrialized countries, this may suggest a need for more recognition of and research on catatonia in this vulnerable population. There was also a nonstatistically significant fall in articles on neuroleptic-induced catatonia, malignant catatonia, and NMS. It is unclear whether this may reflect a stabilization or fall in such cases or, more concerningly, decreasing recognition.

Articles reporting on periodic catatonia, recurrent catatonia, and catatonic relapse have fallen dramatically in the new cohort. This drop is concerning and unexpected, since we have much to learn about these types of catatonia, including their contemporary prevalence. In a 1974 longitudinal study of catatonic schizophrenia, the authors noted "recurrent episodes are the rule rather than exception".[51] Whether this is true for contemporary patients with catatonic schizophrenia and how rates differ with catatonia associated with other conditions are unclear. However, in a 2016 case series of 30 patients with catatonic recurrence and relapse, 63% had schizophrenia, 17% major depressive disorder, 7% bipolar disorder, and 13% general medical conditions.[52] A 1965 review on periodic catatonia with notes regarding a decrease in the number of cases of periodic catatonia at that time "suggests that the great majority of patients suffering from periodic catatonia are helped sufficiently by means of ECT or phenothiazines to escape notice."[53]

It is our experience that recurrent catatonia is commonly encountered and that clinicians often struggle with how to prevent relapses, particularly when it comes to the question of standing benzodiazepines on an outpatient basis.[54] The field would benefit from further studies of this issue, as well as studies on the use of ECT,[55] lithium,[56] clozapine,[57] and other medications to prevent recurrence and relapse.

Of particular concern is a drop in both absolute numbers and proportions in the new cohort of articles reporting findings from animal/*in vitro* studies, genetic/heredity studies, and interventional clinical trials. There also appears to be stagnation in neuroimaging studies in catatonia, which is surprising given how much more easily accessible neuroimaging resources have become since 2003 and the fact that neuroimaging is now used widely to study other psychiatric conditions. To further our insights into the neural underpinnings of catatonia using neuroimaging methods, some authors have called for resting-state neuroimaging studies of catatonia longitudinally within individual cases, as well as comparison neuroimaging studies of patients with varying comorbid conditions, symptoms, and chronicity.[58]

Taken together, the fall in research studies on catatonia suggests that although interest in catatonia may be growing among clinicians, as manifested by an increasing number of case reports, interest in conducting research in this patient population appears to be waning. The reasons for this are not clear although they are likely heavily influenced by the fact that most patients with catatonia are treated in inpatient psychiatric settings, where conducting clinical research is both difficult and rare.[59] Additionally, with ever stricter informed consent requirements for clinical research, enrolling patients with communication difficulties or prominent negativism secondary to catatonia in interventional clinical trials and neuroimaging studies has become more challenging, likely due to the detriment of care improvement for this patient population. Given the small number of interventional clinical trials identified in this study, including only 2 articles reporting results exclusively from randomized, placebo-controlled trials,[59] it is clear that additional clinical trials are sorely needed to better characterize the efficacy, tolerance, and protocols of treatment, as identified by a recent systematic review.[60] Research evidence for treatment modalities other than ECT and lorazepam remains quite limited,[61] and we can only hope that the recent wave of nosological interest in catatonia will be followed by a similar wave of interventional clinical trials for catatonia.

Other complications of the catatonia research efforts, as revealed in a 2011 systematic review,[62] are the different ratings scales for catatonia, each utilizing different but overlapping underlying concepts of catatonia, differing nature and number of included items (signs and symptoms), and differing diagnostic thresholds. The BFCRS appears to be one of the most commonly used in clinical practice and currently seems to be the most used in research, as evident by our finding that the article introducing it in 1996 is the most-cited article on catatonia.[25] Since its development, other rating instruments, including the Kanner scale,[63] have been created with the intention of addressing these shortcomings although they have yet to displace the BFCRS in practice or research. Other important rating scales include the Rogers scale,[64] the Modified Rogers Scale,[65,66] the Northoff Catatonia Rating Scale,[67] and the Braunig Catatonia Rating Scale.[68] Since the various catatonia rating scales have been validated in different patient populations, a better characterization of the conflicting elements of these instruments and their respective strengths could provide psychiatrists with a more refined understanding of them.

Limitations

Primary strengths of this study are the fact that it is the first published bibliometric analysis of the catatonia literature and that it covers more than 55 years, providing valuable insights into how the catatonia literature has changed over generations. Additionally, we believe this to be the first content analysis of the bulk of the catatonia literature, providing valuable novel information on how the characteristics of articles on catatonia have changed before and after 2003.

There are several important limitations to our study. Particular metrics of influence may be seen as problematic or simply unproductive. For example, articles are cited for many reasons, and simply being the most-cited does not necessarily make an article the most influential on our understanding of a topic. Rather, it indicates that an article is somehow significant although this could be due to being controversial or highly criticized rather than being influential. Using citation counts as a measure of "importance" is just as complex of a topic as using other metrics to measure an author's implied influence through other scientometric/bibliometric techniques (i.e., h-index) or a journal's impact (i.e., impact factor). Therefore, rather than choosing to highlight specific authors or institutions with the most article output or citations, we chose to focus on the growth of the catatonia literature and the theoretical and contextual evaluation of this body of work.

Other important limitations of bibliometric studies include errors of commission (inclusion of irrelevant papers) and omission (exclusion of relevant papers), often heavily influenced by the database investigated and search terms employed. To mitigate these risks, we did not select PubMed as our data source due to Web of Science's more robust publication metadata and its built-in tools for basic bibliometric analysis. We also attempted to use broad-enough search terms used in other published systematic reviews on catatonia, designed to capture many articles on catatonia, without risking including articles that are not specifically on catatonia.

CONCLUSIONS

The literature on catatonia is the product of many authors spanning multiple countries, academic institutions, and eras. Encouragingly, it also appears to be experiencing significant growth in recent decades. Using bibliometric analysis and content analysis, we found that since 2003, the proportion of case reports in the catatonia literature has increased, while the proportion of research articles has fallen. The catatonia literature appears to have matured in that it is increasingly inclusive of articles detailing catatonia associated with a general medical condition and substance use. However, drops in the proportions of articles reporting findings from animal/*in vitro* studies, genetic/heredity studies, and interventional clinical trials, along with a stagnation in neuroimaging studies, are causes for concern. With this bird's eye view of catatonia research now available to the field, we hope that it might inspire new lines of inquiry that further our understanding, diagnosis, and treatment of this mysterious malady.

SUPPLEMENTARY DATA

Supplementary data related to this article can be found at https://doi.org/10.1016/j.jaclp.2022.07.002.

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