

Review

Research on dissociative seizures: A bibliometric analysis and visualization of the scientific landscape

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ABSTRACT

Dissociative seizures are a common and often elusive differential diagnosis in epilepsy centers. Considering their high prevalence, long diagnostic delays, and disappointing rates of treatment response, scientific research dedicated to dissociative seizures is surprisingly scarce. In order to chart the scientific landscape of dissociative seizures and to visualize thematic clusters and trends in research, a comprehensive bibliometric analysis was performed. The Web of Science database was examined to identify relevant English language documents from the last half-century. A total of 1751 documents with titles referring to dissociative seizures were identified. Automated textual analysis of all titles and abstracts revealed that research clusters around three major topics: differential diagnosis in epilepsy centers, management and treatment, and psychopathology. Time analysis of term networks revealed that the focus of clinical research has moved from diagnostic procedures to treatment approaches. Furthermore, interest within etiological research is shifting from an emphasis on early life trauma and personality traits to the role of anxiety and emotion regulation. With respect to individual contributing authors, a relatively small network of prolific scientists with a remarkable degree of collaboration emerges. By mapping relevant publications, it becomes evident that dissociative seizures still represent a subject mostly within the realm of neurology and epileptology, with a tendency to settle in the latter domain. This analysis sheds light on an important niche subject and highlights trends in research focus and output.

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1. Introduction

Functional neurological disorders, also known as conversion disorder and formerly as “hysteria”, tend to occupy the “no-man’s-land” between neurology and psychiatry [1]. Neurologists often remain suspicious regarding malingering and leave that distinction for psychiatrists to make, whereas psychiatrists are often wary of unrecognized underlying brain pathology and tend to ask for repeated testing [1,2]. This overall reluctance of both disciplines to fully commit to research and treatment of functional neurological disorders has resulted in a striking scarcity of research and clinical service provision. Dissociative seizures, a prominent form of functional neurological disorder, seem to have fared slightly better. Because of their close semiological resemblance to epileptic seizures (and common co-occurrence), dissociative seizures have long been “adopted” by epileptologists as a diagnosis within their responsibility. This seems inevitable since 25–30% of patients seen in epilepsy centers for suspected refractory epilepsy are found to have dissociative seizures [3]. Still, “nonepileptic” seizures have remained a Cinderella subject among neurologists [2], and it is

not only the epidemiology that would dictate higher scientific engagement—the average diagnostic delay from first dissociative seizure to final diagnosis is still 5–10 years, long-term treatment outcomes are largely disappointing, and the socioeconomic burden of these disorders is considerable [4,5]. An important obstacle to improving care for affected patients is a paucity of evidence-based models of pathomechanism. Recent efforts to elucidate the neural underpinnings of dissociative seizures have not yet resulted in a complete mechanistic understanding [6]. Despite these persisting challenges, however, progress in recognizing and treating dissociative seizures has been made since the introduction of video-electroencephalography (video-EEG) monitoring in 1968 [7]. In order to chart the scientific landscape of the last half-century and to explore clusters and trends of research on dissociative seizures, we conducted a comprehensive bibliometric study on this subject.

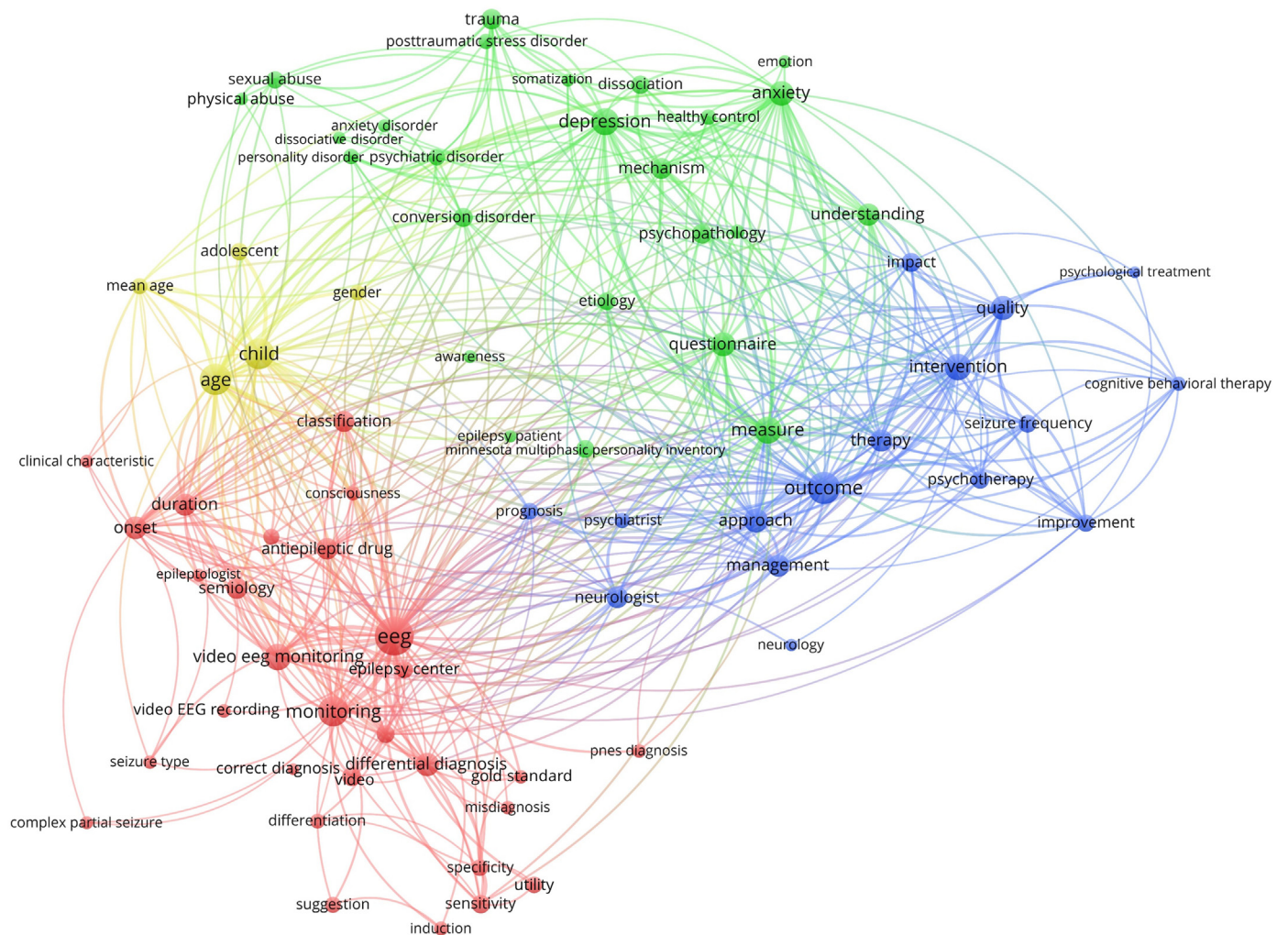
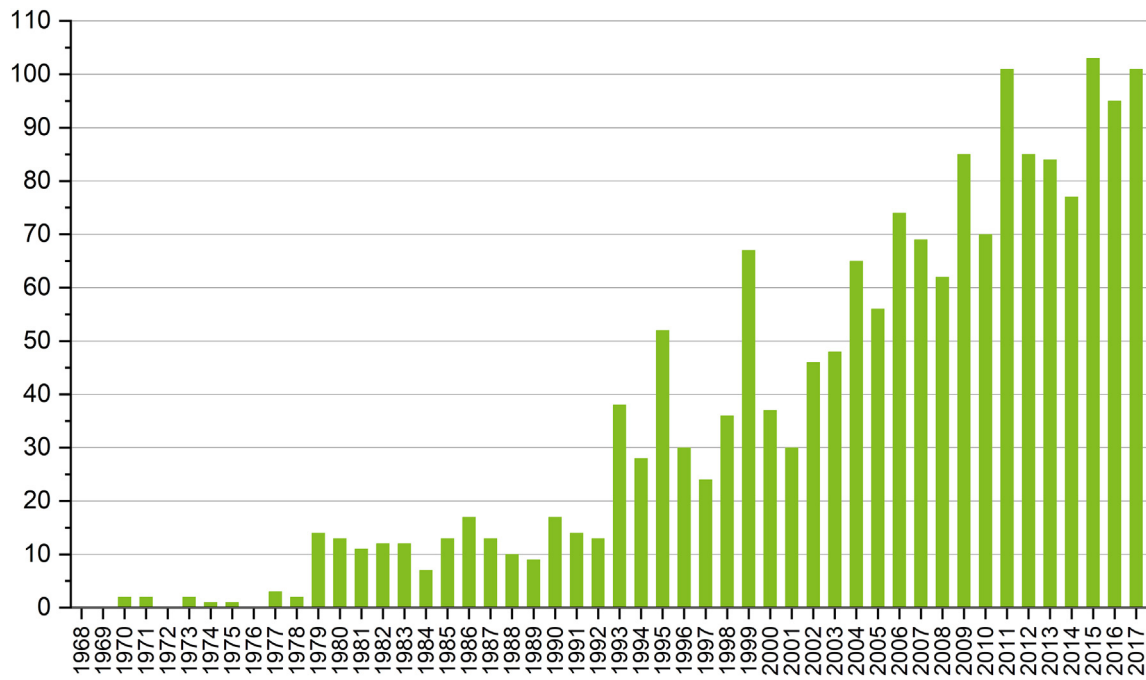
2. Methods

2.1. Data collection

The Web of Science Core Collection database was chosen to identify and extract relevant research documents. The Web of Science is a human-curated database (unlike Google Scholar) which tracks citations

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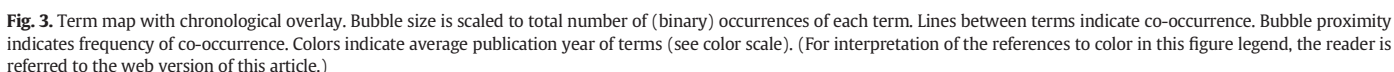
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TI = ("nonepileptic attack" OR "non-epileptic attack" OR "psychogenic attack" OR "nonepileptic attacks" OR "non-epileptic attacks" OR "psychogenic attacks") OR TI = ("nonepileptic event" OR "non-epileptic event" OR "psychogenic event" OR "nonepileptic events" OR "non-epileptic events" OR "psychogenic events") OR TI = ("nonepileptic episode" OR "non-epileptic episode" OR "psychogenic episode" OR "nonepileptic episodes" OR "non-epileptic episodes" OR "psychogenic episodes") OR TI = ((psychogenic OR nonepileptic OR non-epileptic OR dissociative OR hysteric*) AND (seizure* OR convulsion*)) OR TI = (hysteroepilep* OR hystero-epilep* OR pseudoseizure* OR pseudo-seizure*)

they were included. To assess the chronology of research output, the number of publications per year was analyzed for the time period 1968–2017. In order to contextualize the trends in publication output, the compound annual growth rate was calculated based on the change in publication numbers over the fifty-year time period and was then compared with that of all Web of Science documents from the categories “Clinical Neurology” and “Psychiatry” in the same time period. Extracted document (meta)data were imported to VOSviewer version 1.1.6, a software tool for the construction, analysis, and visualization of bibliometric networks [10–12]. All further analyses and visualizations detailed below were conducted using VOSviewer.

Unique terms were automatically extracted from the titles and abstracts of all documents in the dataset and were used to construct a map based on textual data [12]. Terms were counted in a binary fashion, meaning that each term was counted only once per item. A custom “thesaurus” was employed to avoid redundancy (e.g., “behavior” and “behaviour” counted as the same term). The minimum number of occurrences of a term in unique items to be included in the analysis was set to 20. Thus, each identified term received a relevance score. The top 60% of terms according to this score were included in the analysis. Next, all terms were manually inspected, and uninformative general usage terms such as “life” and “reduction” were excluded.



Also, synonyms for dissociative seizures that were identified (such as “NEAD” or “PNES”) were excluded.

2.3. Coauthorship map

A network visualizing authorship statistics in the field of dissociative seizures was constructed using the aforementioned dataset of items. Of the total number of authors, those who have (co)authored at least 10 documents in the dataset were selected for further automatic clustering and network visualization.

2.4. Map of sources

As mentioned above, the dataset was reduced to exclude meeting abstracts since they could skew the network properties in favor of official journals of societies. The network structure of scientific journals in the field of dissociative seizures was explored by creating a map of sources within the dataset that visualizes the relatedness of publication sources based on the number of times they cite each other. A brief custom “thesaurus” was used to group together items in cases when the source has changed names (e.g., *Journal of Epilepsy* was renamed *Epilepsy Research*).

3. Results

3.1. Dataset

A total of 1751 documents from the last half-century with titles referring to dissociative seizures were identified. Fig. 1 shows a steady trend of increasing publication output each year, rising from about 7.5 per year in the first half of the evaluated timespan (1968–1992) to about 62.5 per year in the second half (1993–2017). About half of all documents (863) were published in the last decade (2008–2017). Since there were no published documents in 1968 and 1969, the compound annual growth rate was calculated for 1970–2017 and amounted to 8.90%. For comparison, the compound annual growth rate for all documents in the Web of Science database in the categories “Clinical Neurology” and “Psychiatry” for the same time period was 6.85% and 4.45%, respectively.

3.2. Term map

A total of 14,336 terms were automatically identified from the titles and abstracts of all 1751 items. Of those, 238 terms occurred a minimum of 20 times. The top 60% of those (143 terms) were selected and inspected manually according to automatically assigned relevance scores. Uninformative terms (such as “value”, “none”, or “month”) were

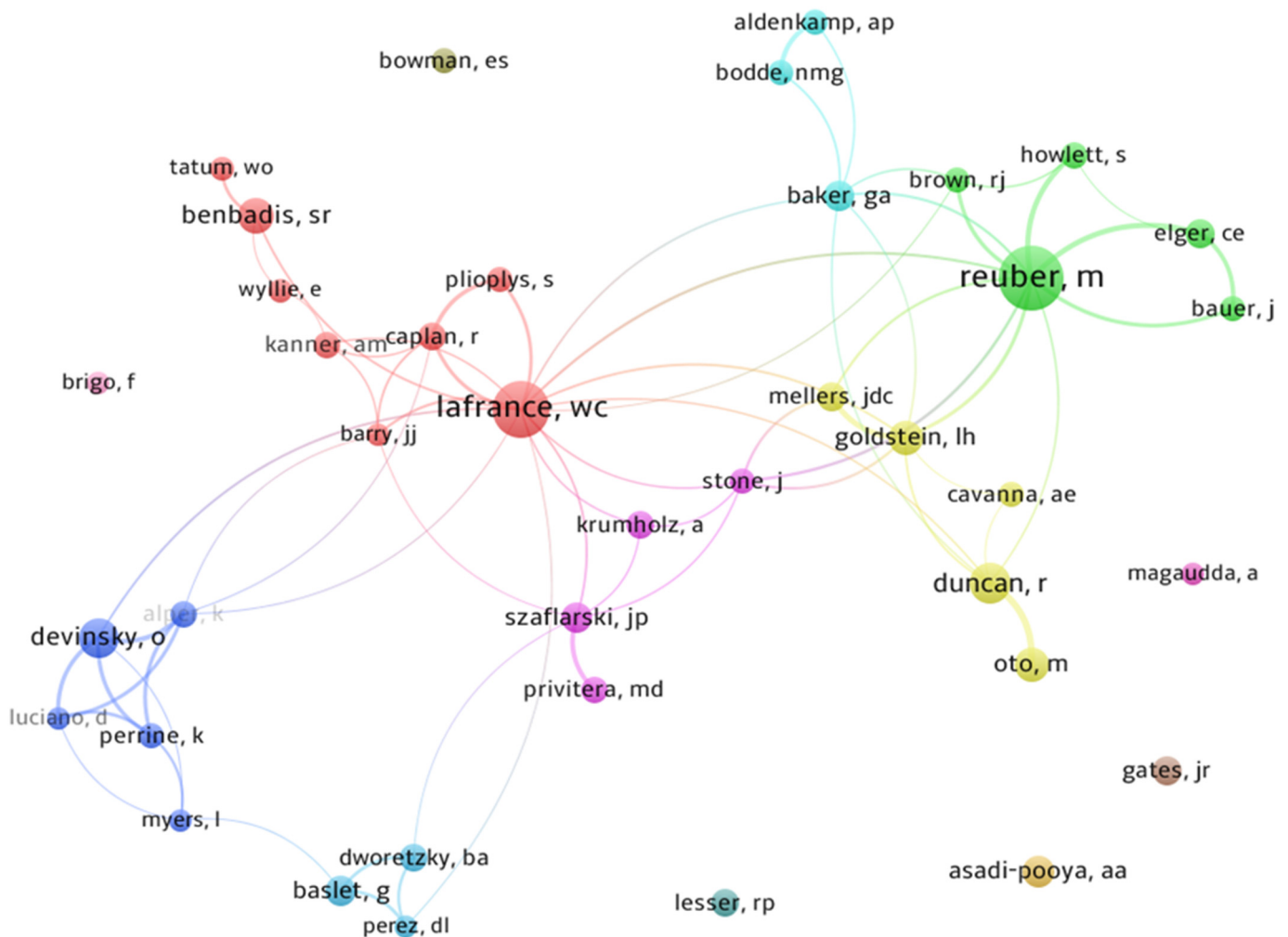


Fig. 4. Authors map. Bubble size is scaled to number of documents published. Links and bubble proximity represent coauthorship. Colors represent clustering based on coauthorship. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

were excluded, as were synonyms of “dissociative seizures”. The remaining 75 terms were analyzed and visualized. Fig. 2 shows the generated term map. Cluster analysis based on term co-occurrence identified three major and one minor color-coded thematic clusters. Fig. 3 shows the same term map color-coded for time (average publication year of term).

3.3. Coauthorship map

A total of 3830 authors were identified. Of those, 39 authors had (co) authored at least 10 documents in the dataset. Fig. 4 charts these 39 authors with automatic color-coded clusters of cooperation based on coauthorship.

3.4. Map of sources

This analysis was performed after excluding all documents with the document type “Meeting abstract” and is, thus, based on 1168 items. Fig. 5 visualizes the network of influential sources (16 journals and one book) that have published a minimum of 8 documents from the dataset. All five major epilepsy journals are represented as well as an influential book of conference proceedings called “Nonepileptic seizures” [13]; several major general neurology journals; and a few psychiatric, psychosomatic, and interdisciplinary journals.

4. Discussion

The bibliometric analysis of 1751 scientific documents relating to dissociative seizures yielded several interesting features of the steadily growing scientific landscape. The term map and network analysis revealed four thematic clusters (Fig. 2). One is mostly concerned with differential diagnosis in an epileptological setting (red) with prominent terms such as “EEG”, “video-EEG monitoring”, “classification”, “specificity”, and “sensitivity”. This reflects a major effort since the 1980s to standardize and optimize the diagnostic process [3], which recently culminated in the formulation of systematic diagnostic guidelines for dissociative seizures issued by the International League Against Epilepsy in 2013 [9]. Nowadays, the gold standard in diagnosing dissociative seizure in an epileptology setting comprises video-EEG monitoring with or without induction techniques [9,14,15]. Another cluster relates to management and treatment, comprising terms such as “seizure frequency”, “outcome”, and “psychotherapy”, as well as, tellingly, both “neurologist” and “psychiatrist”. The chronological overlay (Fig. 3) reveals a shift in the focus of clinical research from diagnostic procedures to treatment. Terms such as “complex partial seizure”, “suggestion”, and “monitoring” have much earlier occurrences on average than terms such as “psychological treatment” and “cognitive behavioral therapy”. A third large cluster (green in Fig. 2) clearly comprises terms relating to psychopathological mechanisms and psychiatric comorbidities with keywords such as “depression”, “anxiety”, and

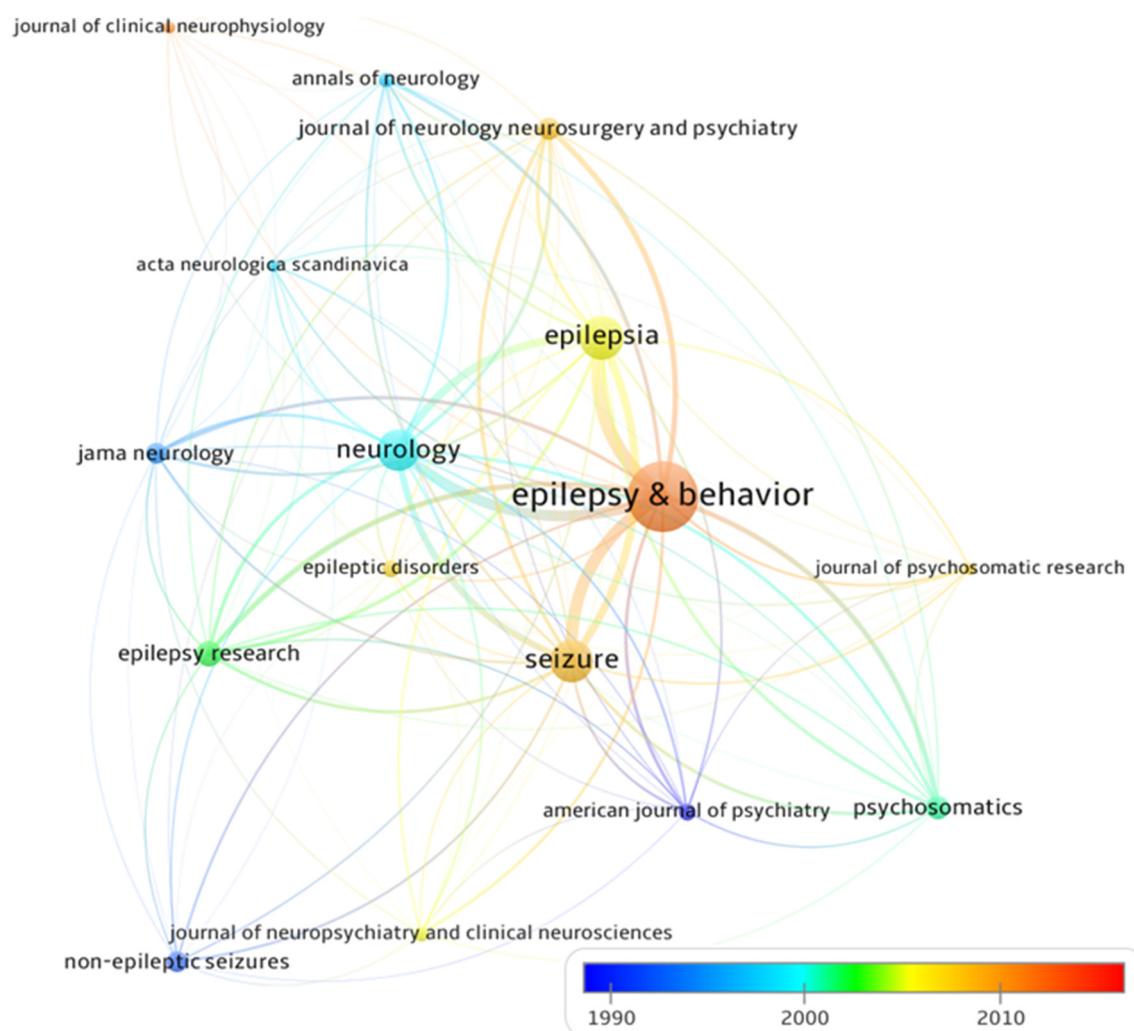


Fig. 5. Network of sources. Bubble size is scaled to number of published documents. Links and bubble proximity indicate citation between sources. Colors represent average publication year of all documents published by each source. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)

“questionnaire”. The time-coded overlay map (Fig. 3) reveals an interesting trend within this cluster. Interest in sexual and physical abuse and personality traits, formerly a focus of etiological inquiry, seems to have waned in favor of posttraumatic stress disorder (PTSD), conversion disorder and depression, and most recently, anxiety and emotion regulation. This reflects a current trend in neuropsychiatric conceptualization that de-emphasizes early life trauma as a direct mechanism (while recognizing it as a risk factor [16]) and places the focus on anxiety and emotion regulation [17]. This is seen in the latest edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), where psychological risk factors such as childhood trauma are recognized as commonly associated with conversion and dissociative disorders but are not considered prerequisite features anymore [18,19]. Similarly, anxiety management and emotion regulation have become central to modern therapeutic strategies [20]. A minor thematic cluster (yellow in Fig. 2) seems to converge around questions of clinical demography with terms such as “mean age” and “gender” but only comprises five terms. Interestingly, recent efforts to localize relevant neural dysfunction with structural and functional imaging techniques [6,21] have not yet registered in this broad automated literature analysis.

Metadata from all documents were used to reveal the most prolific authors and the most impactful sources. The total number of authors associated with the literature is high, but there are only 39 individuals with 10 or more coauthored documents in the dataset. However, the relatively small research community is remarkably well connected. This kind of collaboration and interconnectedness has recently allowed for large multicenter trials on both sides of the Atlantic, which are urgently needed to establish evidence-based treatment strategies [20,22]. The map charting the top 16 sources in the dataset reveals that research on dissociative seizures has been published predominantly in general neurology and epileptology journals. As has been noted previously, *Epilepsy & Behavior* stands out as a leading publication in the field [23], and while it is of note that psychiatric publications are relatively underrepresented, the fact that several decidedly interdisciplinary journals were identified in this search points to a psychiatric awareness of the topic within an interdisciplinary setting.

A bibliometric study such as this one can only provide a rough overview of trends in research and publishing and is limited by several factors. The input data were extracted using an elaborate search string, but might still have included irrelevant or duplicate titles, and might have missed pertinent publications that have unspecific titles. Furthermore, the thematic analysis is based solely on the frequency of term occurrence and co-occurrence without further semantic evaluation. Including meeting abstracts into the textual analysis will also have distorted the data in favor of studies that were published first as abstracts and then as full journal articles. Overall, even though computational algorithms are only beginning to assist with semantic analyses of vast datasets, this simple bibliometric study of the literature on dissociative seizures provides a coherent and insightful sketch of the development and structure of the scientific landscape.

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Conflict of interest

All authors declare no conflict of interest.

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