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ORIGINAL RESEARCH

Knowledge Atlas of Insular Epilepsy: A Bibliometric Analysis

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Objective: In order to determine research hotspots and prospective directions, this work used VOSviewer and CiteSpace to assess the current state of insular epilepsy research.

Methods: We looked for pertinent research about insular epilepsy published between the first of January 2000 and the thirtieth of April 2022 in the Web of Science Core Collection (WoSCC) database. CiteSpace and VOSviewer were used to build a knowledge atlas by analyzing authors, institutions, countries, keywords with citation bursts, keyword clustering, keyword co-occurrence, publishing journals, reference co-citation patterns, and other factors.

Results: A total of 305 publications on insular epilepsy were found. Nguyen DK had the most articles published (37), whereas Mauguière F and Isnard J had the highest average number of citations/publications (39.37 and 38.09, respectively). The leading countries and institutions in this field were the United States (82 papers) and Université de Montréal (40 papers). Authors, countries, and institutions appear to be actively collaborating. Hot topics and research frontiers included surgical treatment, functional network connectivity, and the application of neuroimaging methods to study insular epilepsy.

Conclusion: In summary, the most influential articles, authors, journals, organizations, and countries on the subject of insular epilepsy were determined by this analysis. This study investigated the area of insular epilepsy research and forecasted upcoming trends using co-occurrence and evolution methods.

Keywords: visualization analysis, insular epilepsy, CiteSpace, VOSviewer, bibliometrics

Introduction

Penfield reported the first case of epilepsy treated by insular surgery in 1947,[1] and researchers subsequently became aware of the relationship between the insula and epilepsy. Until the 1950s, Guillaume and Mazars first proposed the concept of insular epilepsy.[2] Subsequently, more researchers devoted themselves to the research field of insular epilepsy. For instance, Blum et al discovered in 1961 that insular and circum-insular lesions might cause cats to have autonomic-psychic experimental epilepsy.[3] Due to the complex and atypical symptoms of insular epilepsy, such that it can mimic temporal, frontal, or parietal lobe onset seizures, making its differential diagnosis very difficult.[4,5] Given the anatomical position of the insula, it is difficult for ordinary scalp electroencephalography (EEG) to record insular discharge.

Furthermore, surgery for insular epilepsy is extremely difficult, making research on insular epilepsy difficult. In 1964, Silfvenius et al[6] reported the presence of epileptogenic foci in the insula in patients undergoing temporal lobectomy. However, as the group with added insula resection failed to improve the incidence of postoperative epilepsy and showed an increased incidence of hemiplegia, research in insular epilepsy encountered a bottleneck. When Isnard et al first identified intracranial insular seizures in 2 cases with atypical temporal lobe epilepsy in 2000, there was some progress.[7] Next, in 2004, Isnard et al[8] used stereotaxic electroencephalography (SEEG) to record insular discharge with simultaneous electrical cortical stimulation and systematically summarized the symptoms of insular seizures. In recent years, with an increased understanding of the insular epilepsy network and the development of imaging technology, micro

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neurosurgery, and SEEG, research in insular epilepsy has progressed. As a result, it is vital to summarize and organize the research on insular epilepsy conducted over the last 20 years.

Using software for correlation analysis, bibliometrics is a quantitative approach for describing and analyzing the dynamics and development of a discipline or study area.[9,10] To conduct bibliometric and visual analyses for this work, we employed the bibliometrics-related software programs CiteSpace and VOSviewer.[11,12] Recent systematic and narrative reviews on insular epilepsy highlight the diagnostic methods and surgical treatments of insular epilepsy.[4,5] Bibliometrics differs from traditional narrative reviews providing readers with the current status and trends of global research in a certain research field, such as research themes and research cooperation. As far as we are aware, there has been no bibliometric study of research on insular epilepsy reported until now. We performed a quantitative study and visualization of cooperative networks (authors, institutions, and countries), co-citation references, published journals, keyword citation bursts, keyword clustering, and keyword co-occurrence in insular epilepsy over the previous two decades. As a result, the current study examines the fundamental, frontier, and hot themes in insular epilepsy research systematically and unbiasedly.

Methods

Sources of Data and Searching Strategy

Data were obtained using powerful search techniques of the Web of Science Core Collection (WoSCC), an expanded version of the Science Citation Index, as illustrated in Figure 1. In the topic field, the following search keywords were entered: "insula epilepsy" OR "insular epilepsy" OR "insula seizures" OR "insular seizures" OR "insula seizure" OR "insular seizure". The period covered was from the January 1,2000 to April 30, 2022; the only language allowed was English, and the only types of papers acceptable were reviews and original research articles. Following the lead of earlier studies,[9] two independent researchers examined the abstracts and titles and the criteria for inclusion were any publications relevant to insular epilepsy. Finally, 305 papers were considered for the subsequent study.

Data Analysis and Visualization

The bibliometric analysis and data visualization were carried out using VOSviewer 1.6.18 and CiteSpace 6.1.R2. First, we constructed a text file called download XXX.txt that contained all of the records we had obtained from WoSCC in plain text. After that, these data were uploaded into CiteSpace and VOSviewer software for bibliometric and visual

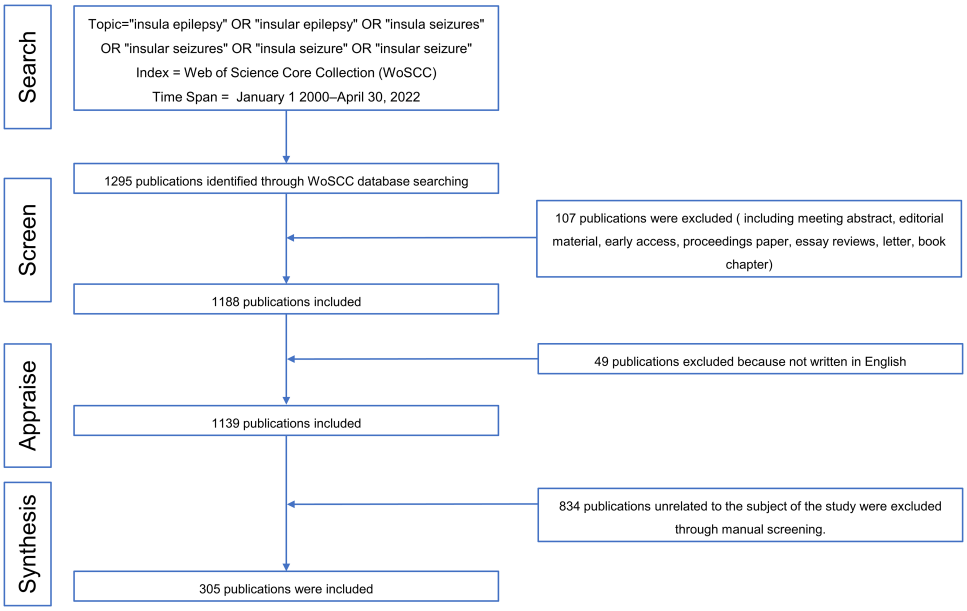

Figure 1 Depicts a flow diagram of the literature's initial screening procedures.

analysis. Drawing visual maps from the analysis of countries, journals, co-authors, co-citation references, co-occurring keywords, and their clusters. CiteSpace tracks keywords with significant historical citation bursts and examine annual publication counts and growth trends to identify research fronts and upcoming trends.

Results

Bibliometric Analysis of Publication Years

Figure 2 displays 305 articles on insular epilepsy publication dates between January 2000 and April 2022. With minor oscillations, the proportion of publications on insular epilepsy has typically increased over time. Before 2008, less than 10 articles were published each year. Since 2009, the number of published papers has risen to more than 10 per year. Each year from 2017 through 2021, more than 20 papers were published. In 2019, 33 articles were published, making it the year with the most articles. Only 6 articles had been published that year as of April 30th, 2022.

Bibliometric Analysis of Journals

In this study, the publication volume of 81 journals was analyzed. First, journals with five or more publications in insular epilepsy research were visualized using VOSviewer (Figure 3). The journals with the most publications were then evaluated. Seven journals produced more than 10 publications annually, totalling 136 papers (44.6%). *Journal of Neurosurgery* (19, 6.2%), *Epilepsy & Behavior* (30, 9.8%), and *Epilepsia* (33, 10.8%) were the top three journals (Table 1).

Countries and Institution Bibliometric Assessment

From the first of January 2000 to the thirtieth of April 2022, only 44 countries participated in insular epilepsy research. First, using VOSviewer, countries with three or more papers were shown (Figure 4). Table 2 provides the top five countries producing insular epilepsy research to further assess high-productivity countries. The USA, France, Canada, China, and Italy were the top 5 nations with articles published. 257 papers from these countries were published in total, making up 84.3% of all publications, the United States having the most (82, 26.9%) published articles. A total of 484 institutions were associated with the authors of the 305 papers. As shown in Table 2, the top five institutions with the most publications were Université de Montréal, Université Lyon 1, Hospital Civils Lyon, Capital Medical University, and Aix-Marseille University. These five institutions published 91 papers, accounting for 29.8% of all articles.The institution with the most publications was the Université de Montréal (40, 13.1%).

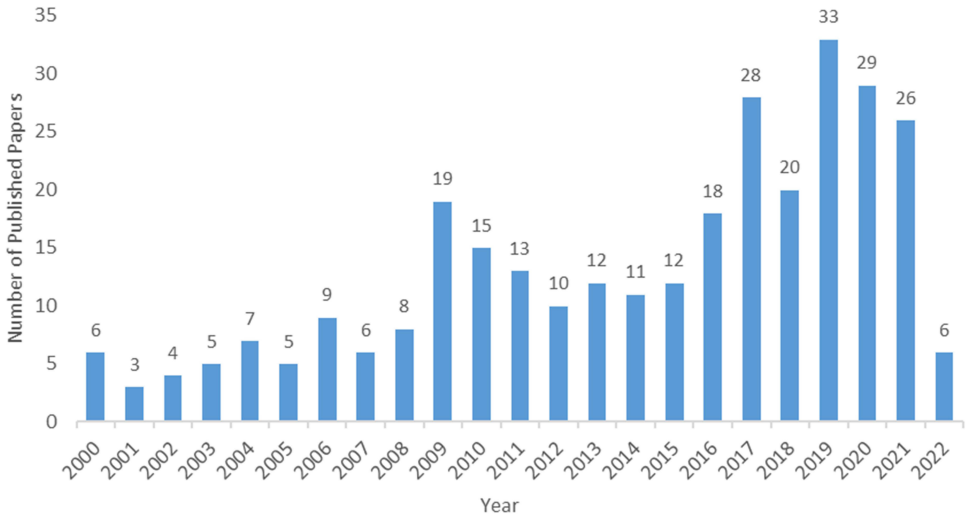

Figure 2 Shows the number of yearly publications changes from the first of January 2000 to the 30th of April 2022.

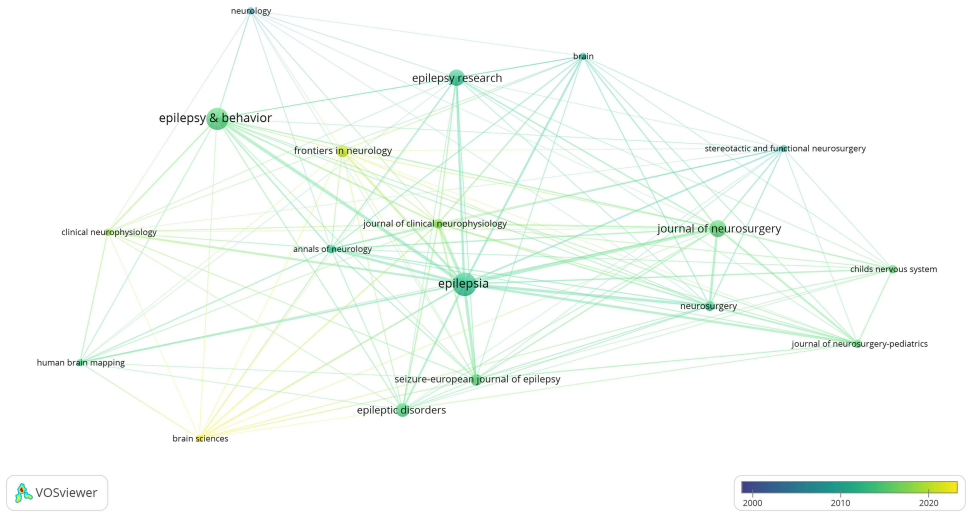

Figure 3 Visualization of the network for the published journals. The number of published papers increases with node size. The node connection lines show the strength of the connections between journals. The node's colour shows how the number of articles published has changed over time.

Co-Authors' Citations Analyses

We used VOSviewer to represent the authors with 5 or >5 publications. Figure 5 incorporates a time factor via overlay representation, with the colour gradient naturally expressing the active status of many researchers in recent times. The connecting lines indicate collaborations, the node colour denotes the time distribution (yearly average of published articles), and the node size shows the number of published papers. We compiled the researchers with more than 10 articles to learn which authors have significantly advanced the understanding of insular epilepsy research. Nguyen DK (37, 12.1%), Bouthillier A (30, 9.8%) and Boucher O (15, 4.9%) were the top 3 leading contributors by the number of publications (Table 3). From 2000 to 2022, the author with the most publications was Nguyen DK, with 37 publications. Bouthillier did not, however, have the highest average publication/citations. The top five researchers with the greatest average number of publications/citations were F. Mauguière, J. Isnard, L. Minotti, P. Kahane, and A. Bouthillier, indicating that these authors have a substantial academic impact in the field of research on insular epilepsy.

Co-Cited References Bibliometric Evaluation

To visualize the 7087 cited references, we used VOSviewer selecting the minimum number of citations to 15, a total of 68 references were used for the co-citation analysis of cited references (Figure 6). [1,5–8,13–39] The top 10 references are listed in Table 4.[1,6–8,13–18] Isnard J. (2004; 127 times), Isnard J. (2000; 105 times), and Nguyen DK (2009; 81 times) were the top three co-cited references.

Table 1 List of the Journals Having More Than 10 Articles in the Field of Research on Insular Epilepsy

Rank	Journal	Publications	Citations	Average Number of Citations Per Publication
1	*Epilepsia*	33	2072	62.79
2	*Epilepsy & Behavior*	30	600	20
3	*Journal of Neurosurgery*	19	537	28.27
4	*Epilepsy Research*	18	335	18.62
5	*Epileptic Disorders*	14	143	10.22
6	*Frontiers in Neurology*	11	87	7.91
7	*Seizure-European Journal of Epilepsy*	11	256	22.27

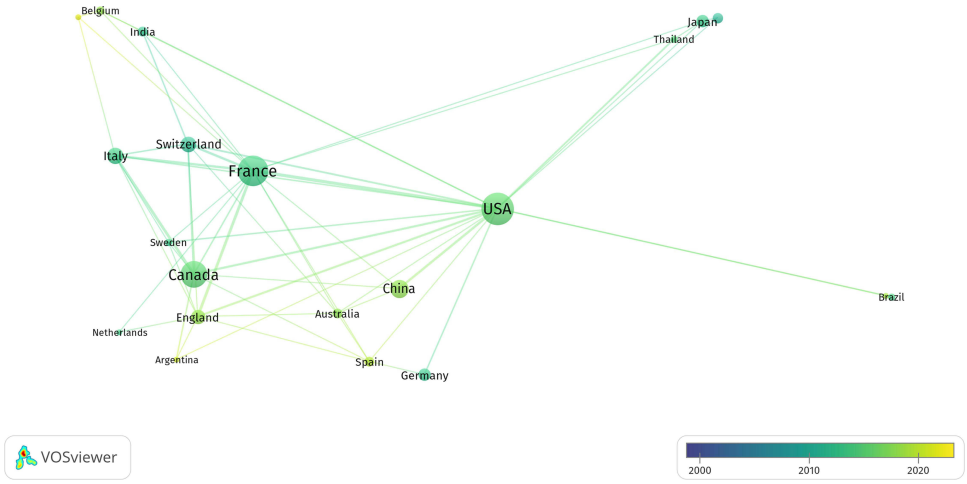

Figure 4 Network visualization of countries. The number of published studies increases with node size. The strength of the connections between nodes represents the relationships between countries. The node's colour displays how the quantity of articles published over time has changed.

Co-Occurring Keywords and Clusters Bibliometric Assessment

VOSviewer was used to draw a keyword co-occurrence network view of the 305 documents. For visualization, we chose 63 important keywords with a frequency of at least 10 (Figure 7). The top 10 co-cited keywords were insula (frequency: 128), epilepsy (frequency: 120), temporal lobe epilepsy (frequency: 83), cortex (frequency: 82), seizures (frequency: 76), surgery (frequency: 69), epilepsy surgery (frequency: 63), insular cortex (frequency: 34), surgical treatment (frequency: 31), and insular epilepsy (frequency: 30).

We utilized CiteSpace to do cluster analysis of co-occurring keywords again to uncover topic groupings of co-occurring keywords. Nine clusters were discovered, with every cluster having an silhouette value of more than 0.7, indicating that the findings are consistent (Figure 8 and Table 5). We used CiteSpace to illustrate the co-clustering timeline view to better understand how these clusters evolved (Figure 9).

Keyword Bibliometric Analysis Using Citation Bursts

Then, we used CiteSpace to visualize the top 10 keywords with the most powerful citation bursts (Figure 10). The figure shows a blue line for the time interval and a red line for the time frame during which the keyword bursts were discovered. The four most recent citation burst keywords occurred in 2017 (insular epilepsy), 2018 (surgery), 2019 (organization, network), and 2020 (lobe epilepsy) and have all lasted until 2022.

Discussion

General Information

This is the very first study to examine the topic of insular epilepsy research using CiteSpace and VOSviewer to identify research hotspots and breakthroughs. Although the concept of insular epilepsy has existed for nearly 70 years, breakthroughs were not achieved until Isnard et al[7] first recorded insular seizures in 2000. Therefore, we searched WoSCC for

Table 2 Lists the Top 5 countries and Organizations Publishing Research on Insular Epilepsy

Rank	Country	Number of Publications	Rank	Institution	Number of Publications
1	USA	82	1	Université de Montréal	40
2	France	73	2	Université Lyon 1	14
3	Canada	55	3	Hospital Civils Lyon	13
4	China	26	4	Capital Medical University	13
5	Italy	21	5	Aix-Marseille University	11

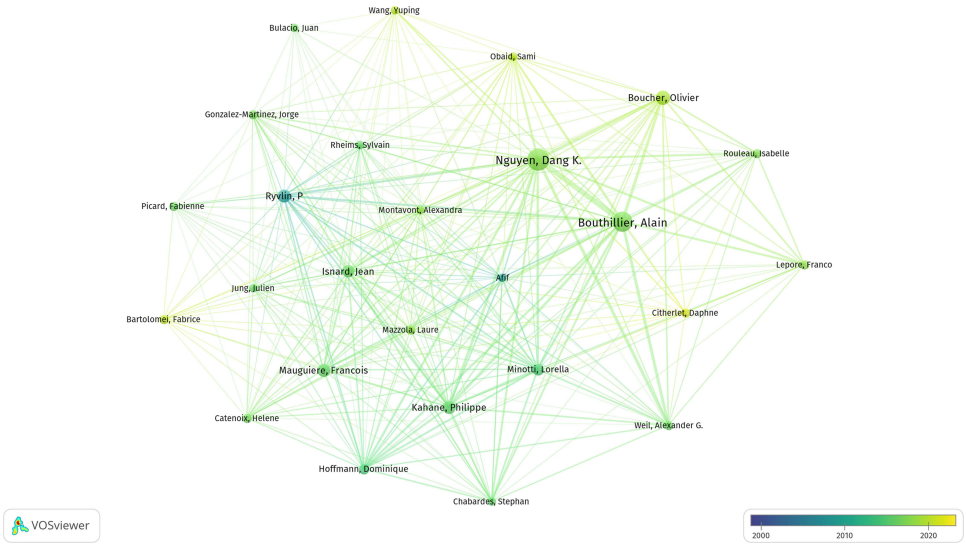

Figure 5 Network visualization of authors. The number of papers published increases with node size. The node connecting lines show the depth of the relationship between authors. The node's colour shows how the number of articles published has changed over time.

articles published from January 2000 to the thirtieth of April 2022. Based on the inclusion criteria, a total of 305 articles were obtained. These 305 articles came from 1409 authors from 484 institutions in 44 countries, were published in 81 journals, and cited 7087 articles from 1172 journals. In general, but with significant oscillations, the number of publications in insular epilepsy studies has grown during the previous two decades. Since 2017, the number of published papers has exceeded 20 per year, which indicates that insular epilepsy is receiving more and more attention from scholars and becoming a new focus in epilepsy research.

Network

Journal Network

Even though an analysis of the published journals cannot describe the current status of insular epilepsy research nor anticipate future research trends, these journals play a crucial role in insular epilepsy research. Thus, these journals are appropriate for creating a theoretical basis and publishing high-quality articles. In our study, 305 articles were published in 81 journals. Further analysis and statistics of the journals revealed that most insular epilepsy research was published in journals belonging to the field of epilepsy and neurosurgery, followed by a small number of comprehensive journals. Most publications were published in *Epilepsia Journal*, with the highest average amount of citations per article. The analysis of papers recently published in this journal revealed topics related to diagnoses, such as the use of magnetoencephalography to assist diagnosis[40] and treatment, including surgical treatment[4,41] and stereotactic thermocoagulation.[42]

Table 3 The Most Influential Authors in the Field of Insular Epilepsy Research

Rank	Author	Number of Publications	Citations	Average Number of Citations Per Publication
1	Nguyen DK.	37	698	18.86
2	Bouthillier A.	30	606	20.2
3	Boucher O.	15	156	10.4
4	Kahane P.	12	351	29.25
5	Mauguière F.	12	476	39.67
6	Isnard J.	11	419	38.09
7	Minotti L.	10	325	32.5

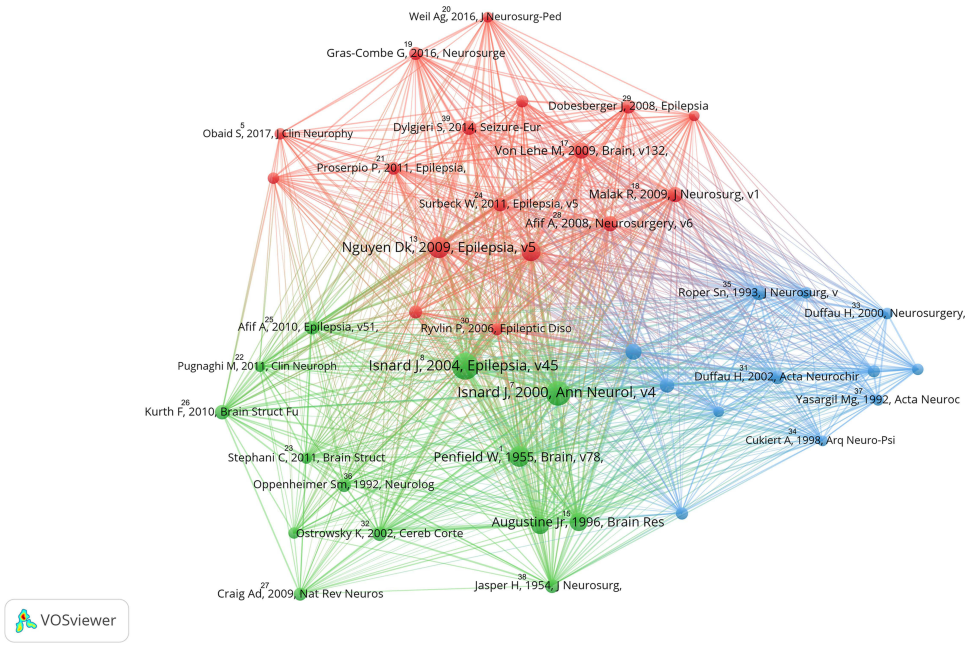

Figure 6 Network visualization of co-cited references. The larger the node, the more citations. The node connecting lines show the association potency of various references referenced in the same article. The node colours represent different categories.

Country and Institution Network

This study examined publishing volumes from 44 different countries and 484 different institutions. In our study, the number of articles in the top five countries and institutions reached 84.3% and 29.8% respectively. These findings show that the top effect is highly significant, most articles are produced by academics from a small number of countries and institutions, and the distribution of countries and institutions producing papers on this subject is relatively uneven.

Author Network

The key research forces and representative academics in insular epilepsy research can be found through the author's cooperation network analysis. Our results also show that the top five authors have a clear cooperative relationship with each other. The two authors with the highest average citations/publication, namely Mauguière F and Isnard J, published breakthrough articles in 2000 and 2004.[7,8] Recent collaborative articles by these two authors mainly focus on electrical stimulation for insular epilepsy or refractory epilepsy[43,44] and the analysis of clinical manifestations of insular epilepsy.[45]

Table 4 Shows the Top Ten Co-Cited References in the Field of Insular Epilepsy Research

Rank	Co-Cited References	Citations
1	Isnard J,[8] 2004, epilepsia, v45, p1079, Doi 10.1111/j.0013–9580.2004.68903.x	127
2	Isnard J,[7] 2000, ann neurol, v48, p614, Doi 10.1002/15318249(200010)48:4<614:: aid-ana8>3.0.co;2-s	105
3	Nguyen DK,[13] 2009, epilepsia, v50, p510, Doi 10.1111/j.1528–1167.2008.01758.x	81
4	Penfield W,[1] 1955, brain, v78, p445, Doi 10.1093/brain/78.4.445	73
5	Ryvlin P,[14] 2006, epilepsia, v47, p755, Doi 10.1111/j.1528–1167.2006.00510.x	62
6	Augustine JR,[15] 1996, brain res rev, v22, p229, Doi 10.1016/s0165–0173(96)00011–2	56
7	Ostrowsky K,[16] 2000, epilepsia, v41, p681, Doi 10.1111/j.1528–1157.2000.tb00228.x	52
8	Von lehe M,[17] 2009, brain, v132, p1048, Doi 10.1093/brain/awp047	46
9	Silfvenius H,[6] 1964, epilepsia, v5, p307, Doi 10.1111/j.1528–1157.1964.tb03338.x	44
10	Malak R,[18] 2009, j neurosurg, v110, p1153, Doi 10.3171/2009.1.jns08807	41

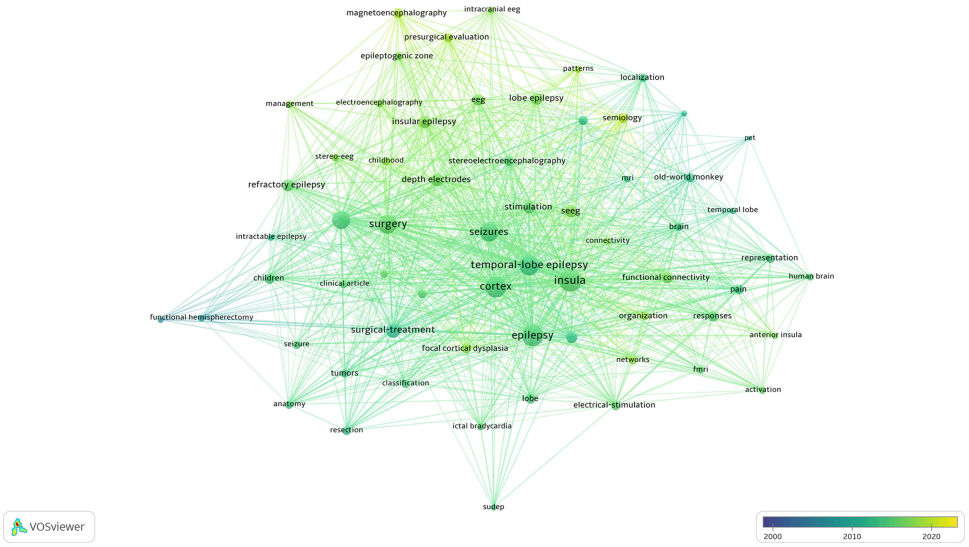

Figure 7 Network visualization of co-occurring keywords. The larger the circle node, the more frequent of an occurrence. The intensity of the association between the frequency of occurrence of several keywords in the same article is shown by the node connecting lines. The node's colour displays how the frequency of occurrence has changed over time.

Co-Cited References Network

Analysis of the cooperative network of co-cited references can reveal the basis of an existing research field. Isnard J. (2004; 127 times), Isnard J. (2000; 105 times), and Nguyen DK (2009; 81 times) were the top three co-cited references. As noted above, the two articles by Isnard J et al in 2000 and 2004 were breakthroughs. Nguyen DK's 2009 article clarified the complexity and diversity of clinical manifestations of insular epilepsy.[13]

Research Frontiers and Hot Topics in Insular Epilepsy

Keywords reveal the essential nature of an article. In bibliometric analysis, terms with a high frequency are utilized to discover research hotspots and frontiers. Through keyword clustering analysis, keyword co-occurrence analysis, and a keyword clustering timeline map that illustrates changes in research hotspots and keywords with citation bursts, we

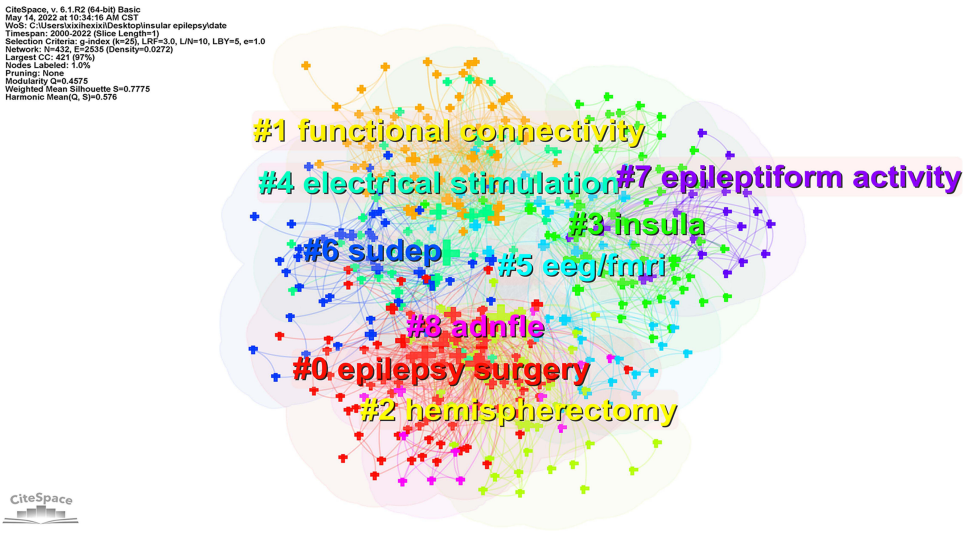

Figure 8 Visualization of the keywords cluster analysis.

Table 5 Keyword Cluster Analysis in the Field of Insular Epilepsy Research

Cluster-ID	Silhouette	Label	Included Keywords (Top 5)	Mean (Year)
0	0.709	Epilepsy surgery	Seizure; epilepsy surgery; surgery; surgical treatment; insular epilepsy	2012
1	0.753	Functional connectivity	Functional connectivity; activation; response; clinical manifestation; monkey	2011
2	0.797	Hemispherectomy	Temporal lobe epilepsy; children; childhood; classification; anatomy	2009
3	0.768	Insula	Brain; complex partial seizure; hippocampal sclerosis; MRI; emission computed tomography	2007
4	0.768	Electrical stimulation	Cortex; epilepsy; electrical stimulation; lobe; astrocytoma	2009
5	0.781	EEG/fMRI	Network; abnormality; fMRI; connectivity; brain activity	2013
6	0.795	Sudep	Stimulation; heart rate variability; ictal bradycardia; cardiac arrhythmia; epileptic seizure	2009
7	0.961	Epileptiform activity	Insular cortex; lesion; microinjection; anterior; ampa receptor antagonist	2003
8	0.91	Adnfle	Localization; basal ganglia; hyperkinetic seizure; frontal lobe; focal seizure	2010

investigated the hotspots and frontiers of insular epilepsy research in this article. Our findings suggest that clusters 0, "epilepsy surgery", 1, "functional connectivity", and 5, "EEG/fMRI", maybe hot issues and possibilities in insular epilepsy research.

Epilepsy Surgery

Surgery for insular epilepsy is difficult because the insula is situated in the deep part of the Sylvian fissure and interacts with the surrounding brain regions, blood arteries, and other structures.[46,47] As a result, surgical treatment of insular epilepsy has always been a hot topic and frontier in insular epilepsy research. When insular epilepsy was first proposed, insular cortectomy was applied. However, this surgical method did not improve outcomes and had high complications due to technical limitations, causing it to be gradually abandoned.[2,6]

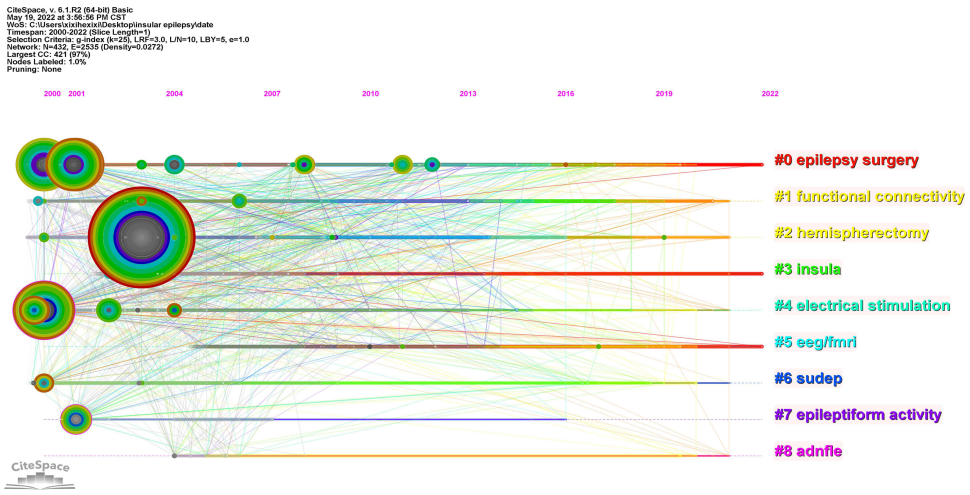

Figure 9 Shows a timeline picture of co-clustering from 2000 to 2022. The node's size and colour represent the overall number of references and specific time slices. Two articles are co-cited in one article and are shown by different coloured lines. The clustering subjects for hot and non-hot topics over time.

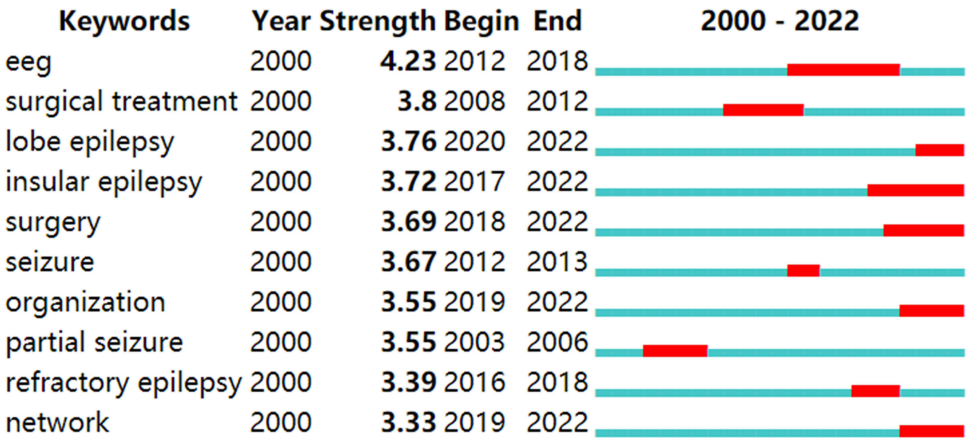

Figure 10 Shows the top ten terms (keywords) with the most citation bursts. Blue line for the time interval and red line for the time frame during which the keyword bursts were discovered. The bold text refers to the start and end time and intensity of the keywords burst.

The development of novel surgical techniques, electrophysiological monitoring, and navigation tools over recent decades has greatly improved the outcomes after insular corticectomy.[18,48] It is believed that partial or complete resection of the insula can improve the prognosis of insular epilepsy.[18,49] For patients with magnetic resonance imaging (MRI)-negative insular epilepsy, precise monitoring and implantation of invasive EEG — especially SEEG — are needed to explore the epilepsy-causing network so that the epilepsy area can be resected in a targeted manner.[50–52]

Alternative surgical modalities for insular resection include SEEG-guided radiofrequency thermocoagulation (RFT),[42,53–55] MRI-guided laser ablation (laser interstitial thermal therapy (LITT)),[56–58] gamma knife radiosurgery,[54,59] and responsive neurostimulation.[60] Interestingly, when we collated the data, we found that although scholars have continued to research traditional craniotomy insular cortextomy, such as bipolar electro-coagulation with cortextomy,[61] more scholars are focusing on minimally invasive procedures, especially LITT.[62–66]

Functional Connectivity

It is well known that seizures can disturb brain networks and lead to connectivity disturbances.[67] As the insula has various functions and forms extensive functional network connections with almost all other brain regions, the manifestations of insular epilepsy are complex and diverse and may even mimic other focal epilepsies. Some scholars have recently begun to study complex networks of functional connectivity in insular epilepsy, and this topic has gradually become a frontier of insular epilepsy research. By analyzing interictal high-frequency oscillations (HFOs) using magnetoencephalography (MEG), Yin et al[68] found that patients with insular epilepsy showed alterations in the insula-based connectivity network. They also reported that alterations in this network might be responsible for interictal brain dysfunction in patients with insular epilepsy. Rachidi et al[69] recently summarized the application of direct cortical stimulation (DCS) to the insula, mapping the functional network connections of the insula to help understand where insular seizures start and how they propagate. Zhao et al[70] showed that insular epilepsy could broadly disrupt the brain metabolic network using fluorine-18-fluorodeoxyglucose positron emission tomography (18F-FDG-PET) imaging. Additional MatLab tools have been developed to study and analyze functional connectivity networks in insular epilepsy.[71]

EEG/fMRI

Given the anatomical position of the insula, general EEG and MRI cannot be used to effectively study insular epilepsy. In parallel with the development of EEG and MRI technology, especially SEEG and functional MRI (fMRI), research on insular epilepsy has made breakthroughs. Recently, Peltola et al[52] used SEEG to analyze the different symptoms of anterior and posterior insular seizures, thereby helping determine the actual onset zone of insular seizures. Insular epilepsy can be further subdivided into insulo-opercular epilepsy, opercular epilepsy, and insular cortex epilepsy, as shown using SEEG combined with PET.[51] As a non-invasive neuroimaging method, fMRI plays a crucial role in the study of insular epilepsy. Recently, some scholars have used fMRI to find structural connectivity changes in insular

epilepsy.[72] Due to the complexity of insular epilepsy, it is often necessary to combine EEG/SEEG, fMRI, MEG, etc., in practical applications.[54,73] Given continuous development in auxiliary examination technology, EEG/fMRI has always been a hot topic and frontier in insular epilepsy research.

Limitations

There are several limitations to this study. First, data were gathered from a single database. Although WoS encompass various journals, it is impossible to include all journals containing insular epilepsy research. Thus, our analysis of data may not be comprehensive enough. Second, our search terms may not be perfect enough to retrieve all publications in the research field.And also only two article types and English language articles were included in our study, which, to some extent, makes the analysis incomplete. Finally, while bibliometric analysis employing professional software is objective, various authors' perspectives and assessments of the same content may differ, possibly resulting in human errors.

Conclusion

The understanding of insular epilepsy is still lacking, and it is a very complex problem for clinical and basic study. Through bibliometric analysis, this study tracked the advancement of insular epilepsy research from 2000 to 2022. We selected significant publications, authors, journals, and nations using 305 papers we downloaded from WoS and further examined the research network. According to our findings, we selected minimally invasive surgery, functional connectivity, and MEG, SEEG, and fMRI applications as hot issues and prospects in insular epilepsy exploration.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors report no competing interests.

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