

# Worldwide Productivity and Research Trend of Publications Concerning Spontaneous Intracranial Hypotension: a Bibliometric Analysis

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## Abstract

**Background and Aim:** Spontaneous intracranial hypotension (SIH) is a highly misdiagnosed disease. The main reason is the lack of awareness. This study sought to summarize the current status of research in this field via a bibliometric analysis.

**Materials and Methods:** The publications were searched in the Web of Science Core Collection (WoSCC) database. Data analysis and visualization were carried out using the cooperative network and cluster analysis of R and Cite Space software.

**Results:** A total of 618 publications from 2008 to 2021 were included in this study. The number of publications has a growth trend. The United States contributed the most articles, and Schievink WI is the most prolific author. Mayo Clinical ranked the first compared with other institutions. American Journal of Neuroradiology has published the most articles in this field. The top 3 clusters of reference co-citation are “delayed recurrence”, “cerebrospinal fluid-venous fistula”, and “floating dural sac sign”. The top 3 clusters of keyword co-appearance are “spinal fluid leak headache”, “symptom duration”, and “cerebral venous thrombosis”. The strongest citation burst of keywords is “CSF-venous fistula” and “paraspinal vein sign”.

**Conclusions:** This study analyzes the research progress of SIH, and the current research mainly focuses on CSF-venous fistula. The bibliometric analysis of SIH is expected to help researchers effectively explore the disease.

**Key words:** spontaneous intracranial hypotension ; bibliometric ; research progress ; cite space

## Abbreviations:

SIH : spontaneous intracranial hypotension

CSF : cerebrospinal fluid

WoSCC: Web of Science Core Collection

SCI-E : Science Citation Index Expanded

HDCT : heritable disorders of connective tissue

MRI : magnetic resonance imaging

LP : lumbar puncture

CT : computed tomography

EBP : epidural blood patches

## Introduction

Spontaneous intracranial hypotension (SIH) is secondary to leakage of cerebrospinal fluid (CSF) at the spinal level and the resulting loss of CSF volume[1]. The most often symptom is headache, especially in an upright position, accompanied by other symptoms such as vomiting, neck stiffness, tinnitus, dizziness, hearing disturbances, etc. [2]. The variability of signs and symptoms and low awareness of the disease led to delayed diagnosis or even misdiagnosis [3, 4]. Despite not generally being considered a life-threatening disease, the incidence of SIH is 5 per

100,000 and may be underestimated[5]. With the delay of the disease and the increase in misdiagnosis rate, SIH may lead to many long-term disabilities and, in a few cases, may also lead to decreased consciousness and coma [6]. Exploring and summarizing the SIH research trend is of significance to those who want to carry out this research, because it allows more people to have a deeper understanding of the SIH.

Bibliometrics uses statistical methods such as cooperative network and cluster analysis to analyze books, articles, and other publications, especially in terms of scientific content. Bibliometric mapping visualizes academic output as a parameter of publication and citation information in specific fields. It allows data to be presented in ways that make relationships more understandable and provide researchers with relatively macro information [7]. It has been widely used in clinical disease research [8, 9]. However, so far, there is still a lack of data on the bibliometric analysis of SIH.

We conducted a bibliometric analysis of SIH-related literature in the past 20 years, explored research trends and frontiers, tracked cooperation and networks, and provided the latest insights and discoveries.

## Materials and Methods

### Data Collection

We searched for publications in spontaneous intracranial hypotension derived from the Web of Science Core Collection (WoSCC) through the Science Citation Index Expanded (SCI-E). The search strategy was TS=(spontaneous intracranial hypotension) AND language =(English) AND Document Type=(Article, Review) AND Year Published=(2008-2021). All the search results records, including title, author information,

keyword, abstract, reference, etc., were exported in TXT format for analysis.

### Data Analysis

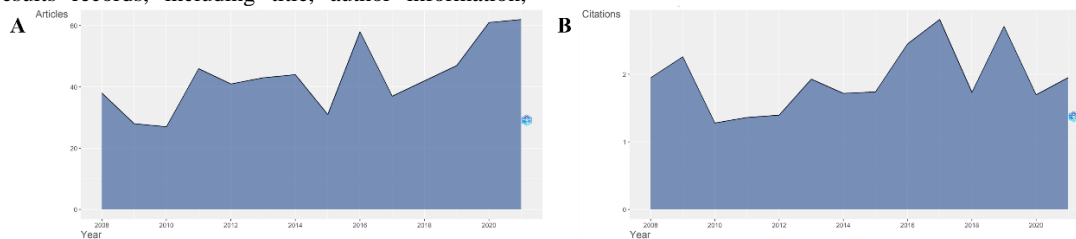
The row files were analyzed by R software (4.1.3) and Citespace software (6.1.R2). The “bibliometrix” R package was used to summarize the primary information, country scientific production, the cumulate occurrence of articles published in journals, and the Three-Fields Plot.

Citespace software (6.1.R2) was used to explore collaboration networks between countries, authors, institutions, journals, references, and keywords. CiteSpace parameters included were as follows: time slicing (2008 JAN–2021 DEC), years per slice (1), links (strength: cosine, scope: within slices), selection criteria (g-index: k = 25), pruning (pathfinder, pruning the merged network), cluster labels (log-likelihood ratio), minimum duration (2), and snap to centroids (0).

## Result

### General overview

A total of 618 publications about spontaneous intracranial hypotension from 2008 to 2021 were included in this study. As shown in Figure 1A, the overall number of articles issued shows an upward trend. The average article citations per year are stable at about 2 (Figure 1B). The record count reached 69, accounting for 11.165% in 2021. The COVID-19 pandemic might have caused fluctuations in production. There are 519 articles and 99 reviews sources from 204 journals.

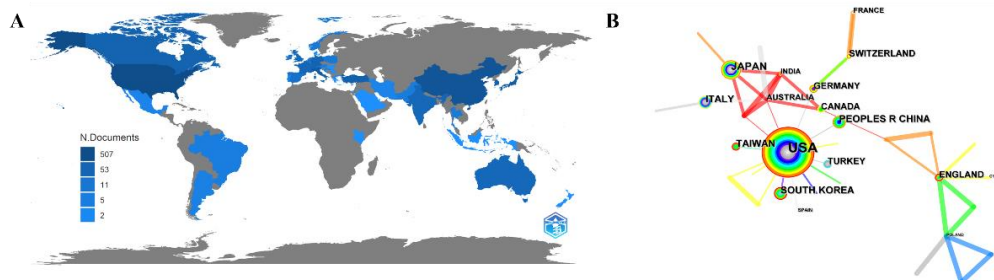


**Figure 1:** (A) Annual scientific production; (B) Average article citation per year

### Countries analysis

A country scientific production map is shown in Figure 2A. The United States contributed the most with 519 publications, followed by China (226 publications), Japan (129 publications), Italy (123 publications), and

Turkey (76 publications). In terms of cooperation between countries, the scientific collaboration was mapping in Figure 2B. There were 47 Nodes and 63 Links. The United States has extensive collaboration with others around the world. International cooperation between other countries needs to be further strengthened.



**Figure 2:** (A) Country-specific production. Dark blue = high productivity. Grey = no documents; (B) Country cooperation network map. The size of each node represents the number of publications published by the country, and the thickness of each link represents the strength of the cooperative relationship between two countries.

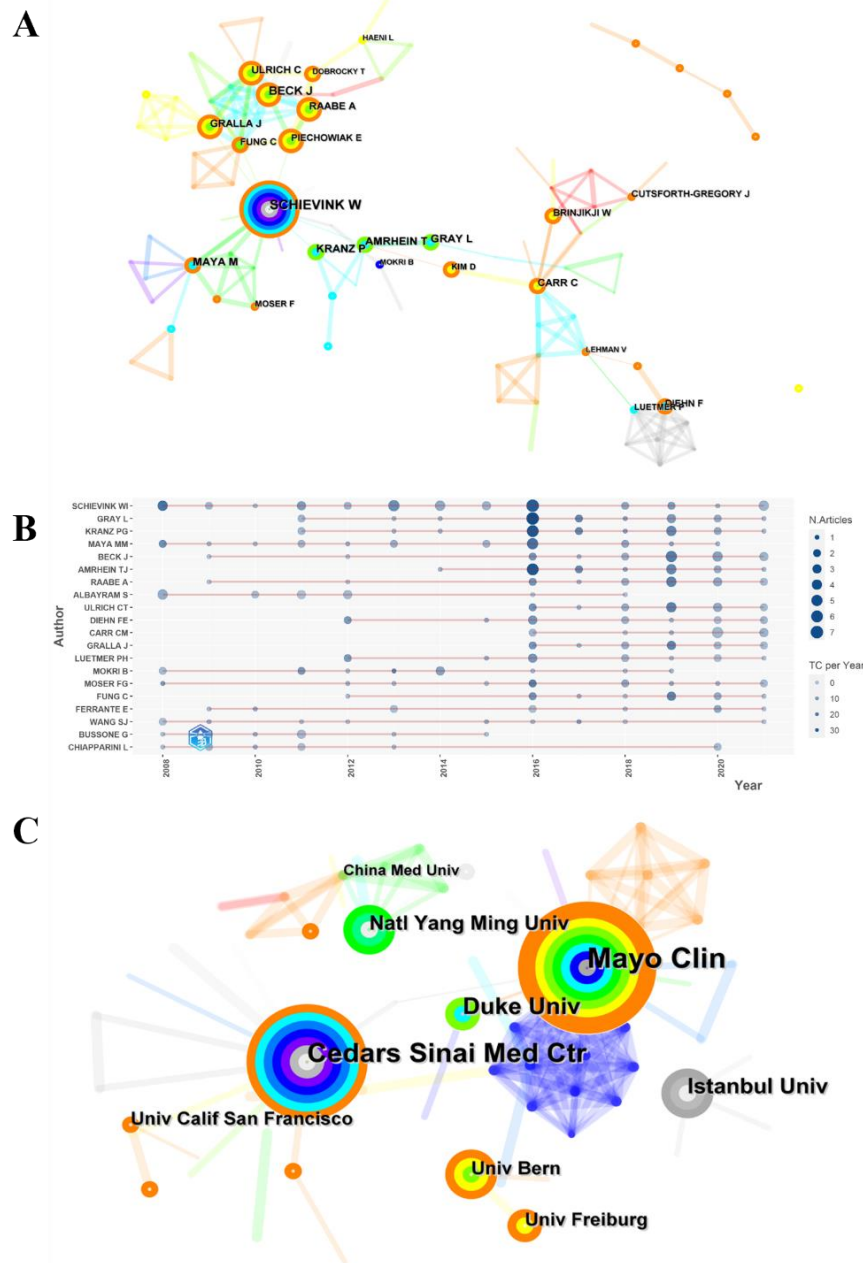
### Authors and Institutions Analysis

The cooperation network between authors is shown in Figure 3A. There were 421 Nodes and 688 Links, and the results showed that Schievink WI

has the most publications with 41 and a centrality score of 0.05. We also visualized the top 20 authors' production over time, which highlights how these authors have been active from 2008 to 2021 (Figure 3B). The top 5 active authors were Schievink WI, Gray L, Kranz PG, Maya MM, and

Beck J. since 2014, Amrhein T and Ulrich C have become active researchers in the field of SIH. In terms of the institutions, there are 376 institutes involved in the field with 324 links (Figure 3C). Mayo Clinical ranked the first with 54 publications, followed by Cedars-Sinai Medical Center (41 publications), Duke University (23 publications), Istanbul University (16 publications), and Natl Yang-Ming University (15

publications). There was a certain degree of cooperation among some institutions, but collaboration was relatively decentralized, which was consistent with the low centrality scores of the institutions. The analysis showed that only Mayo Clinical had a score of 0.02, and Cedars-Sinai Medical Center ad a score of 0.01.



**Figure 3:** (A) Author cooperation network map. The size of each node represents the number of publications published by the author, the thickness of each link represents the strength of the cooperative relationship between two authors; (B) Top-authors' production over time. The size of each node represents the number of publications published by the author, and higher blue intensity represents higher citations; (C) Institution cooperation network map. The size of each node represents the number of publications published by the author, and the thickness of each link represents the strength of the cooperative relationship between two institutions.

**Journals analysis**

A total of 204 journals Participated in the publication of articles in this field (Figure 4A). The top 5 journals in terms of publication volume are

the American Journal of Neuroradiology(41 publications), World Neurosurgery (39 publications), Headache (35 publications), Cephalalgia (22 publications), and Journal of Neurosurgery (20 publications). The

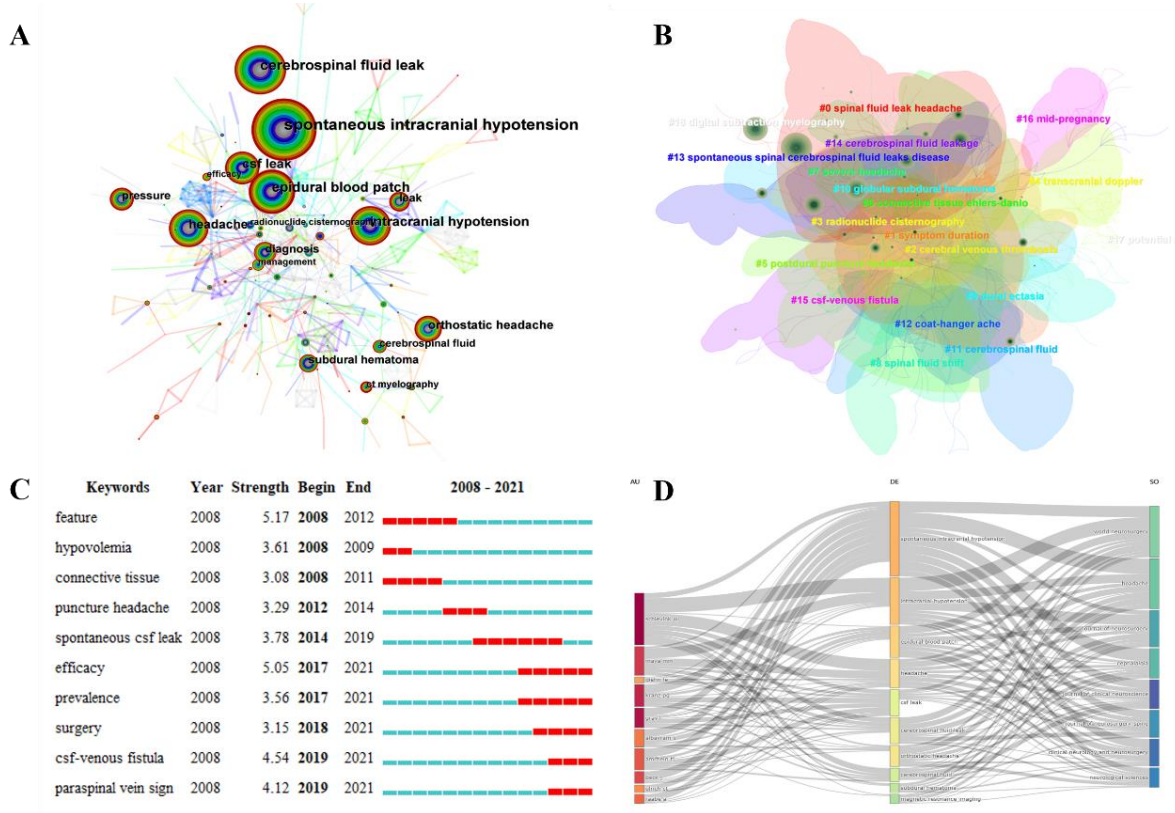


represent more recent references. A straight line in the same horizontal position indicates the set of all clustered references belonging, and the cluster label is located at the line's rightmost end; (D) Top 20 References with the Strongest Citation Bursts.

**Keywords analysis**

There were 446 keywords and 2562 links constructed by Citespace (Figure 6A). The word “spontaneous intracranial hypotension” occurred 364 times, followed by “cerebrospinal fluid leak”(204 times) and “epidural blood patch”(171 times). The keywords cluster is shown in Figure 6B, Modularity Q = 0.7693 and Mean Silhouette = 0.9171. The largest cluster size is “spinal fluid leak headache” among 19 clusters, followed by “symptom duration” and “cerebral venous thrombosis”. The top 10 keywords with the strongest citation bursts indicated that those

study findings of the references were well known in this field (Figure 6C). We found that “paraspinal vein sign”, “CSF-venous fistula”, “prevalence”, “surgery”, and “efficacy” were the most recent burst high-citation keywords. At last, we displayed a Three-Fields Plot to analyze further the hotspots in this field(Figure 6D). We used Left: Authors, Middle: Keywords, and Right: Journey, restricting the number of items to 10 to find out the affiliation of those items. As we expected, Schievink WI made a great contribution to this field, the most important keyword was spontaneous intracranial hypotension, and most articles were published in World Neurosurgery.



**Figure 6:** (A) Co-cited reference map. The size of each node represents the citation frequency of the document; (B) Clustering analysis of keyword co-appearance. Keywords of the same type are clustered in the same color block; (C) Top 10 Keywords with the Strongest Citation Bursts; (D) Three-field plot of classification by author, keyword, and journal.

**Discussion**

Through bibliometric analysis, 618 past medical publications on SIH derived from WoSCC were included in this study for the first time, summarizing the research trends in this field in recent years, promoting the cooperation between researchers from different teams, and providing reference and guidance for future researchers.

We found an increasing trend in articles published in this field since 2008, with an explosive peak in 2016. This may be related to the widespread acceptance and application of non-targeted epidural blood patch therapy. According to the countries analyzed, the United States published the most articles, a total of 238, far more than other countries or regions. Mayo Clinic, the world-leading medical institution, has published the most articles in terms of the author's institution. Journal analysis shows that the American Journal of Neuroradiology published the most articles in this field, And there are many high-cited articles published in this journal. The

analysis of authors showed that Schievink WI from Cedars-Sinai Medical Center had published the most periodicals. In the past 15 years, he has published 41 articles, shared his new ideas with the world almost every year, and made outstanding contributions to the development of this field. However, the authors' centrality scores were low. This phenomenon also exists in the analysis of country and institutional, meaning that multi-country and multi-institution collaborations have not been well carried out in this field.

Co-citation reference analysis means that if two documents appear together in the bibliography of the third cited document, the two documents form a co-citation relationship. These high-cited publications comprehensively describe and summarize SIH and provide the latest research results in epidemiology, pathophysiology, diagnosis, and treatment [1, 10-13]. SIH is caused by CSF volume reduction due to spinal dural tears [14]. Due to the characteristics of the brain, when the CSF volume is reduced, there is less fluid available to buffer the skull in the

brain, resulting in a series of symptoms [15]. The decrease of CSF volume in upright posture has a greater impact on the brain, which is the cause of positional headache in patients with SIH. There is evidence that the potential weakness of the spinal dura mater has been found in a considerable number of cases. Some heritable disorders of connective tissue (HDCT) diseases have been associated with SIH [16]. In addition, there are many cases related to calcified discs and bone spurs, which can tear the dura in the front of the spinal cord[17]. The early diagnosis of SIH relies on lumbar puncture (LP) to measure CSF opening pressure, but the pressure in most patients falls in the normal range. Hence, the diagnostic value of LP is quite limited[18]. Later studies have found that magnetic resonance imaging (MRI) of the brain can obtain some useful information (“SEEPS”, subdural fluid collections, enhancement of the meninges, engorgement of venous structures, pituitary hyperemia, sagging of the brain)[19]. However, these signs are not typical in approximately 20% of cases. Further examinations included spinal MRI, dynamic X-ray computed tomography (CT), digital subtraction, and intrathecal gadolinium-enhanced MR myelography. Noninvasive spinal MRI is usually the initial imaging examination[20-22]. According to the above examination results, There are four types of CSF leaks (Type 1 is caused by a dural tear located ventral (type 1a) or posterolateral spinal cord (type 1b). Type 2 is associated with simple (type 2a) or complex (type 2b) meningeal diverticula. Type 3 is CSF-venous fistulas. Type 4 is of indeterminate origin)[13, 23]. For those patients with mild symptoms, simple measures including bed rest and water supplements may alleviate the symptoms[24]. The most common initial treatment is non-targeted epidural blood patches (EBP) when symptoms are persistent or significant. During this process, The patient's venous blood is injected into the spinal canal of the epidural space to help fill the breach[25]. These procedures can be repeated several times. Emergency treatment is needed in case of serious complications, such as coma or large subdural hematoma [26]. After treatment, most patients have a good prognosis[27], although some patients have recurrent or persistent symptoms and disabilities.

Combined with the citation bursts and keyword bursts, the study of SIH can be divided into three stages. The first stage was before 2013. we had a preliminary understanding of SIH diagnosis. The second stage was from 2013 to 2016. This stage mainly explored the treatment of patients with SIH, and EBP was widely adopted. The third stage is after 2016. The research focuses primarily on the imaging characteristics of SIH because it can help us understand the process of disease occurrence and development, also help us distinguish different types of disease, and guide further treatment.

For the first time, our research conducted a systematic and multi-dimensional analysis of SIH-related publications in an objective way, which can be used as a comprehensive guide for clinicians and scholars engaged in this field and guide the future research direction. But this study inevitably has some limitations. We only retrieve data from the WoSCC database, which leads to incomplete literature collection. On the other hand, the literature retrieved was limited to those published in English, leading to some linguistic bias in the study results.

## Conclusions

This study focuses on the worldwide research results of SIH and analyzes the research hotspots, Frontiers, and development trends in this field. The number of SIH-related productions has increased steadily, which indicates that this field has attracted more and more attention. Especially “CSF-venous fistulas” is the most concerned topic in the current research. Finally, it must be emphasized that there is an urgent need to increase the collaboration between countries and institutions for in-depth basic analysis to optimize the treatment of patients with SIH and improve clinical efficacy.

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