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R E V I E W

# Trends and Hotspots in the Health Economics Evaluation of Chronic Fatigue Syndrome

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**Objective:** To explore the research trends and hotspots of health economics evaluations of patients with chronic fatigue syndrome.

**Methods:** To explore the research trends and hotspots of health economics evaluations of chronic fatigue syndrome, 180 articles published between 1991 and 2024 were visualized and analyzed via CiteSpace 6.3 software. R3 and VOSviewer1.6.20 and R4.3.3. The content includes annual publication volume, journal distribution, author country, publishing organization, author collaboration, citation analysis, and keyword analysis in 7 aspects.

**Results:** Fewer studies have evaluated the health economics of individuals with chronic fatigue syndrome in China and abroad, Chinese studies are especially rare, and research results in the UK are mostly found in other countries. Moreover, cooperation and linkages between institutions, as well as between authors, are not yet strong.

**Conclusion:** The hotspot of health economics evaluation methods in this field is cost-effectiveness analysis, and the hotspot of diagnosis and treatment methods is cognitive–behavioral therapy. We also found that chronic fatigue syndrome may also have a strong potential association with depression from the perspective of health economics. Health economic evaluations of multiple treatments should be conducted simultaneously to increase attention to this field and provide a reference basis for low-cost and high-quality diagnostic and treatment programs.

**Keywords:** chronic fatigue syndrome, myalgic encephalomyelitis, health economics, CiteSpace, visual analysis, bibliometrics

## Introduction

Chronic fatigue syndrome (CFS), formerly known as myalgic Encephalomyelitis (ME), is a complex disease characterized by fatigue symptoms that cannot be relieved even after rest, recurrent episodes or lasting for six months or more, accompanied by symptoms such as sore throat, decreased attention, and muscle pain.[1,2] The incidence of chronic fatigue syndrome ranges from 0.1% to 0.7%, and many people are unable to work or can only engage in part-time work. Patients require long periods of rest and occupy social and other time. It is estimated that individuals with chronic fatigue syndrome abroad lose nearly $20000 per year. Patients face high economic pressure and are not conducive to socio-economic development.[3,4]

The epidemiology of chronic fatigue syndrome (CFS) has been a topic of interest in the medical community for several decades. Manu et al reported that the average age of individuals with CFS was 38.1 years, with an average duration of fatigue of 6.9 years at the time of entry into the study. The authors highlighted the need for further research to determine whether CFS is a distinct entity or a variant of psychiatric illness.[5] Bearn et al emphasized the use of the term chronic fatigue syndrome (CFS) following international consensus due to the complexity of the neurobiological aspects associated with this condition.[6] As a result, most of the research on chronic fatigue syndrome has been based on epidemiologic research, and little has been done in terms of biology or pathology.

In the real world, in addition to direct physiological or biological studies of a disease, the evaluation of interventions is also very important.[7] Health economics evaluations play a critical role in informing decision-making processes about health care interventions and policies and explore the use of economic evaluations in different contexts to assess the cost-effectiveness and impact of interventions.[8] As early as 1996, studies comparing differences in employment status, self-reported disability, number of medical visits, types of services received, and other diagnoses among patients with chronic fatigue, chronic fatigue syndrome (CFS), fibromyalgia (FM), and comorbidities of CFS and FM revealed that patients with chronic fatigue, regardless of whether they met the diagnostic criteria for CFS or FM, had significant individual occupational impacts and lower employment rates, indicating a potentially significant economic burden.[9] Peter Thomas et al subsequently evaluated the clinical effectiveness and cost-effectiveness of group-based cognitive–behavioral therapy for the management of fatigue symptoms in patients with multiple sclerosis (MS) through a multicenter randomized controlled trial (RCT), which revealed that fatigue severity and disease were the main objective factors.[10]

However, while the methods of health economics evaluation implemented in patients with chronic fatigue syndrome vary, no single study has summarized them and explored their multifaceted details. Therefore, an overview of the health economic evaluation of patients with chronic fatigue syndrome is necessary. On the basis of bibliometric studies, this study summarizes the cutting-edge hotspots of chronic fatigue syndrome in health economics with the expectation that it will provide ideas for accurately estimating the cost of chronic fatigue syndrome and provide insights into the development of targeted rehabilitation programs and the design of related studies.

## Methods

Bibliometrics, a subfield of informatics, employs mathematical and statistical techniques to analyze literature systems and their bibliometric traits both qualitatively and quantitatively, offering an efficient and accessible approach to research analysis.[11] This methodology has gained broad application across diverse academic fields.[12] Moreover, bibliometric data, particularly when visualized via tools such as VOSviewer and CiteSpace, can vividly illustrate the focal points and developmental trajectories within research domains.[13]

### Research Design

The research design involves a systematic literature review, commencing with a broad retrieval of documents from WOSCC, PubMed, and Scopus, with a focus on English-language articles and reviews from 1991–2024. The initial corpus is then refined by removing duplicates and irrelevant entries, leading to a curated set of 180 documents. Bibliometric analysis follows, examining the trend of global publications; assessing the contributions of countries, institutions, and authors; and analyzing journals, keywords, and references to identify the focal points and evolution in research.

### Data Collection

For data selection, we chose the Web of Science Core Collection (WoSCC), PubMed, and Scopus databases. The Web of Science Core Collection (WoSCC) is highly valuable because it contains carefully curated and complete literature data covering titles, abstracts, keywords, references and citations, making it the resource of choice for scientometric analysis.[14] PubMed is a free search engine maintained by the National Library of Medicine (NLM) that provides access to citations and abstracts from biomedical and life sciences literature. The database includes more than 32 million citations for biomedical literature from MEDLINE, life science journals, and online books dating back to the 1950s. PubMed encompasses a wide range of scientific fields, including but not limited to clinical medicine, nursing, dentistry, veterinary medicine, healthcare systems, and clinical trials, as well as aspects of biomedicine, such as cellular biology, genetics, physiology, and public health. Users can search keywords, authors, journal names, or Medical Subject Headings (MeSH) terms to quickly and accurately find the literature information they need. Additionally, PubMed links to full-text providers and other related databases, offering a powerful resource to support research, education, and clinical practice.[15] Scopus is known for its scope and user-friendly interface, covering a wider range of literature. The incorporation of Scopus databases into a visualization study can expand the content of the analysis.[14,16]

We developed different search formulas on the basis of the categories of different databases (Figure 1). The search formulas we use are as follows:
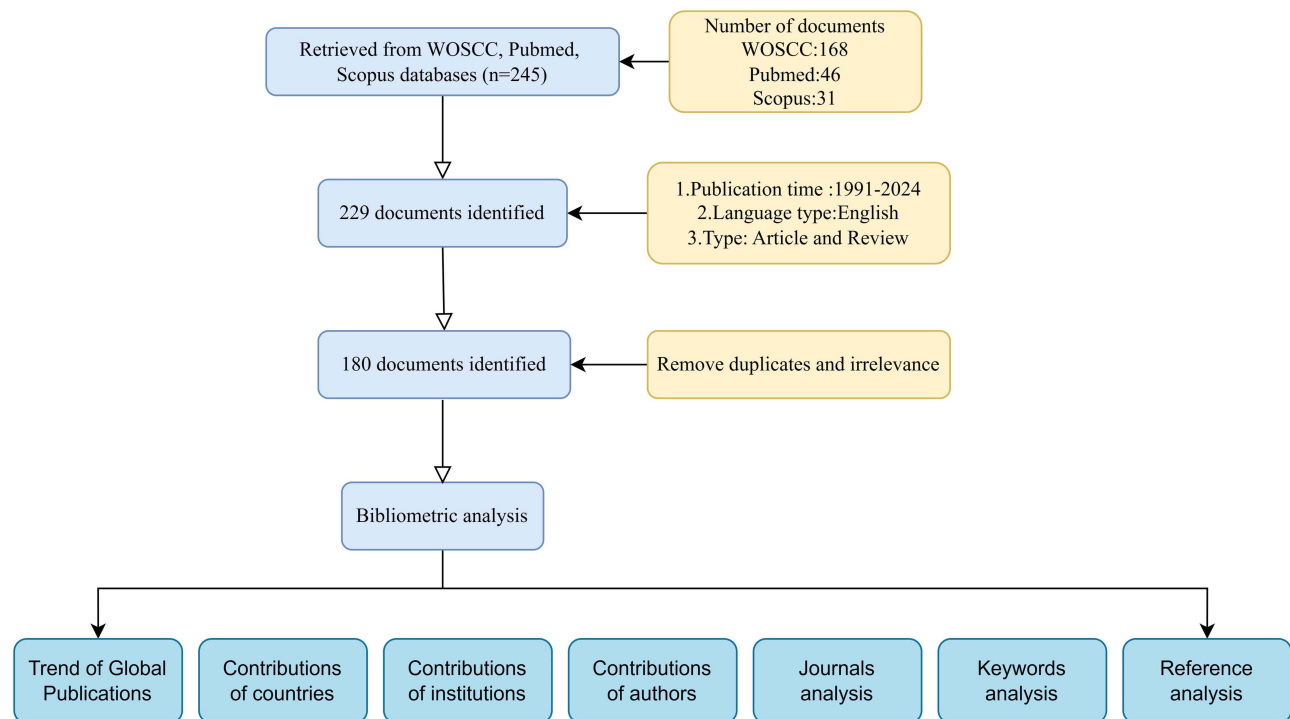
**Figure 1** Flowchart for selection and analysis of publications.

When searching at WOSCC, the search formula is as follows:

(TS=(Chronic Fatigue Syndrome) OR TS=(Chronic Fatigue Immune Dysfunction Syndrome) OR TS=(Systemic Exception Intolerance Disease) OR TS=(Myalgic Encephalomyelitis) OR TS=(persistent fatigue disorder) OR TS=(unexplained chronic fatigue)) AND (TS=(health economics) OR TS=(Medical economics) OR TS=(Healthcare economics) OR TS=(Health care economics) OR TS=(public health economics) OR TS=(Health services economics) OR TS=(Medical cost analysis))

When searching PubMed, the search formula is as follows:

(("Chronic Fatigue Syndrome") OR ("Chronic Fatigue Immune Dysfunction Syndrome") OR ("Systemic Exertional Intolerance Disease") OR ("Myalgic Encephalomyelitis") OR ("persistent fatigue disorder") OR ("unexplained chronic fatigue")) AND (("health economics") OR ("Medical economics") OR ("Healthcare economics") OR ("Health care economics") OR ("public health economics") OR ("Health services economics") OR ("Medical cost analysis"))

When searching Scopus, the search formula is as follows:

(ABS (Chronic Fatigue Syndrome) OR ABS (Chronic Fatigue Immune Dysfunction Syndrome) OR ABS (Systemic Exertion Intolerance Disease) OR ABS (Myalgic Encephalomyelitis) OR ABS (Persistent Fatigue Disorders) OR ABS (Unexplained Chronic Fatigue)) AND (ABS (Health Economics) OR ABS (Medical Economics) OR ABS (Healthcare Economics) OR ABS (Health Care Economics) OR ABS (Public Health Economics) OR ABS (Health Services Economics) OR ABS (Medical Cost Analysis))

# Results

## Temporal Distribution Graph of the Literature

We have plotted the trend of articles published over the years, and the results are displayed as follows. The graph illustrates the number of articles published over time, with the x-axis representing the year and the y-axis representing the number of articles. The data points in the graph show a trend toward an increasing number of articles published over time, with a significant increase in the 2000s and even greater increases in the 2010s and 2020s (Figure 2).
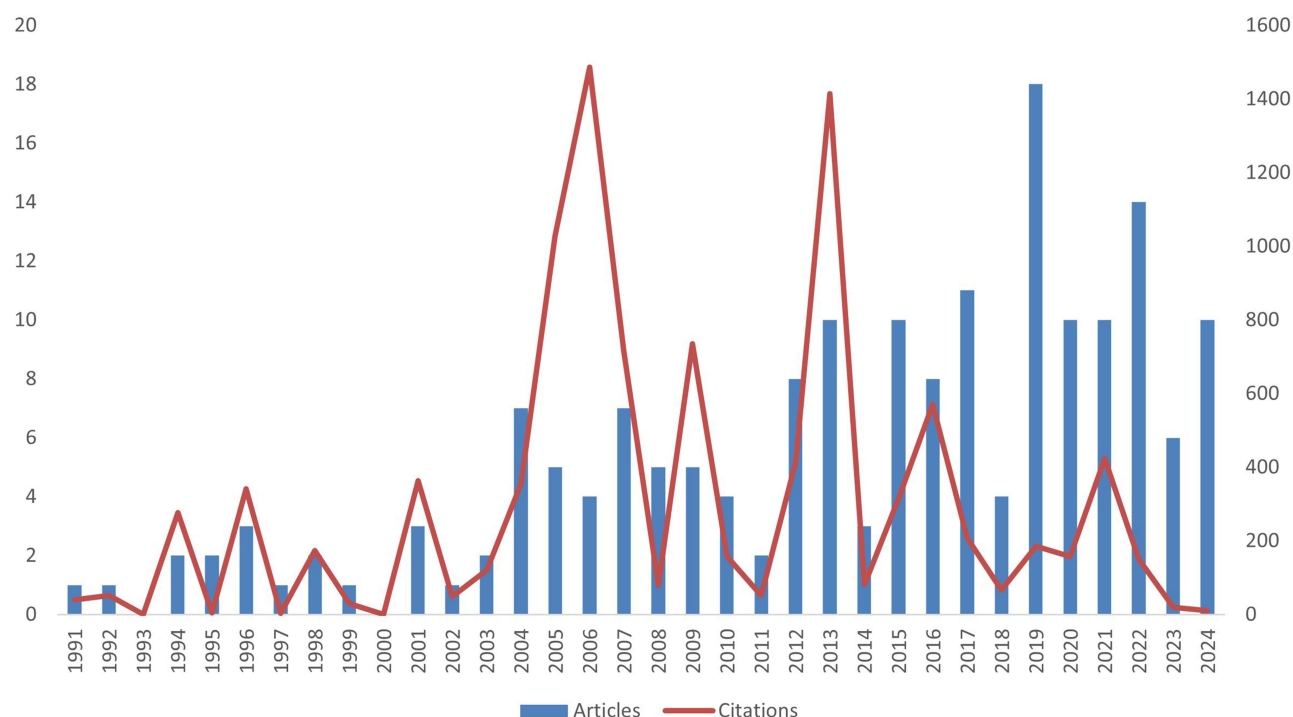
**Figure 2** Number of publications and citations per year.

The time distribution chart shows the ebb and flow of articles published over the years on the health economic assessment of chronic fatigue syndrome (CFS). Initially, from 1991–1994, the chart shows a low and stable number of articles, indicating that very little research was conducted during this period. From 1995–1998, the number of articles began to rise, indicating a growing interest in the field. From 2002–2004, the upward trend continued with steady growth, indicating a period of sustained growth in research activity. However, this growth stabilized between 2005 and 2007 as the number of publications leveled off, suggesting a temporary pause in the upward trajectory. A second significant increase in research activity occurred between 2008 and 2013, with an even more pronounced increase in the number of publications, underscoring the high level of interest in the study of the health economics of CFS during this period. This peak was reached in 2014, representing the highest annual number of publications in the observed time period. After the peak, the chart shows a decline from 2015–2018, indicating a decrease in the number of annual publications. This downward trend continues through 2018, after which the number of articles picked up from 2019 through 2024, albeit at a modest rate of growth, indicating a gradual recovery in research interest and peak levels and suggesting that the 2014 peak remains the most significant surge in research output on the topic over the timeframe analyzed.

## Trend of Global Publications and Evolution of Categories

The trends and hotspots in the health economics evaluation of chronic fatigue syndrome (CFS) are reflected through a comprehensive analysis of global publications and the evolution of research categories. The field has witnessed significant growth in scholarly output, with a diverse array of journals contributing to the discourse. The data presented in Table 1 and Figure 3 provide a detailed overview of the most influential journals and the interconnections among them, highlighting the central role they play in shaping the research landscape.

(Table 1) shows the top 10 most influential journals in the health economics evaluation of CFS, as measured by the number of documents published, citations received, and total link strength. The "Archives of Disease in Childhood" leads with a modest number of documents but a considerable citation count, indicating its significant impact. "BMC Health Services Research" and "BMC Neurology" follow closely, with "BMC Neurology" standing out for its high citation count, suggesting a substantial influence on the field. Notably, "Clinical Infectious Diseases" has an exceptionally high

**Table 1** Top 10 Most Influential Journals in the Field

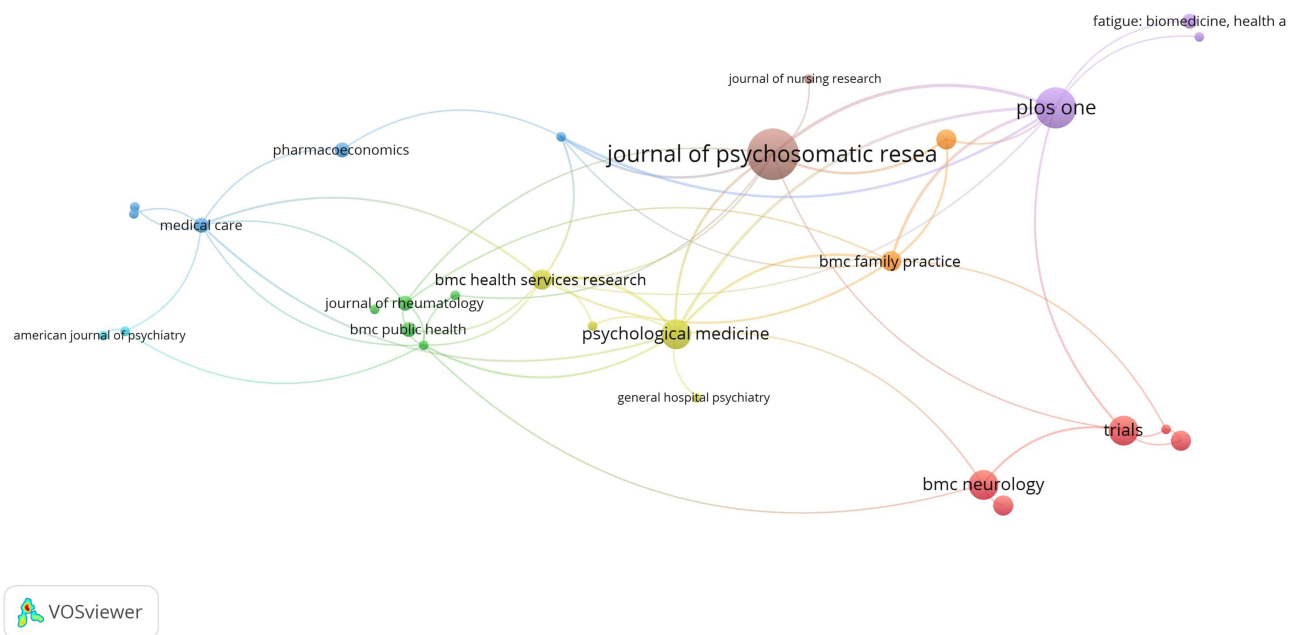| Id | Source | Documents | Citations | Total Link Strength |
|---|---|---|---|---|
| 9 | Archives of disease in childhood | 3 | 37 | 1 |
| 13 | BMC family practice | 3 | 32 | 12 |
| 14 | BMC health services research | 3 | 74 | 11 |
| 15 | BMC medical education | 2 | 0 | 0 |
| 18 | BMC neurology | 5 | 276 | 4 |
| 20 | BMC public health | 2 | 17 | 2 |
| 21 | BMJ open | 3 | 51 | 1 |
| 23 | British journal of general practice | 3 | 58 | 7 |
| 28 | Clinical and preventive medicine | 2 | 2 | 0 |
| 29 | Clinical infectious diseases | 2 | 1355 | 0 |

citation count, which may be attributed to a few highly impactful publications, despite a low total link strength indicating limited interconnectivity among its cited works.

(Figure 3) visually represents the interlinking of journals issuing publications on CFS health economics evaluation. It illustrates the network of citations and references among the journals, providing insight into the collaborative and intellectual relationships within the research community. The graph likely shows "BMC Neurology" and "BMJ Open" as central nodes owing to their high citation counts, suggesting that they are frequently referenced by other works. The interlinking pattern reveals the pathways through which knowledge is shared and built upon, emphasizing the importance of collaborative efforts in advancing the field.

## Distribution of Countries/Regions

The geographical distribution of research efforts in the health economics evaluation of chronic fatigue syndrome (CFS) reveals a global interest with a notable concentration of activity in certain regions. The data from Figure 4 and Table 2 provide insights into the leading countries in terms of publication volume and their collaborative efforts within the field.

(Figure 4) illustrates the global distribution of publications related to the health economics of CFS, highlighting both single-country publications (SCPs) and multiple-country publications (MCPs). This visual representation shows that the



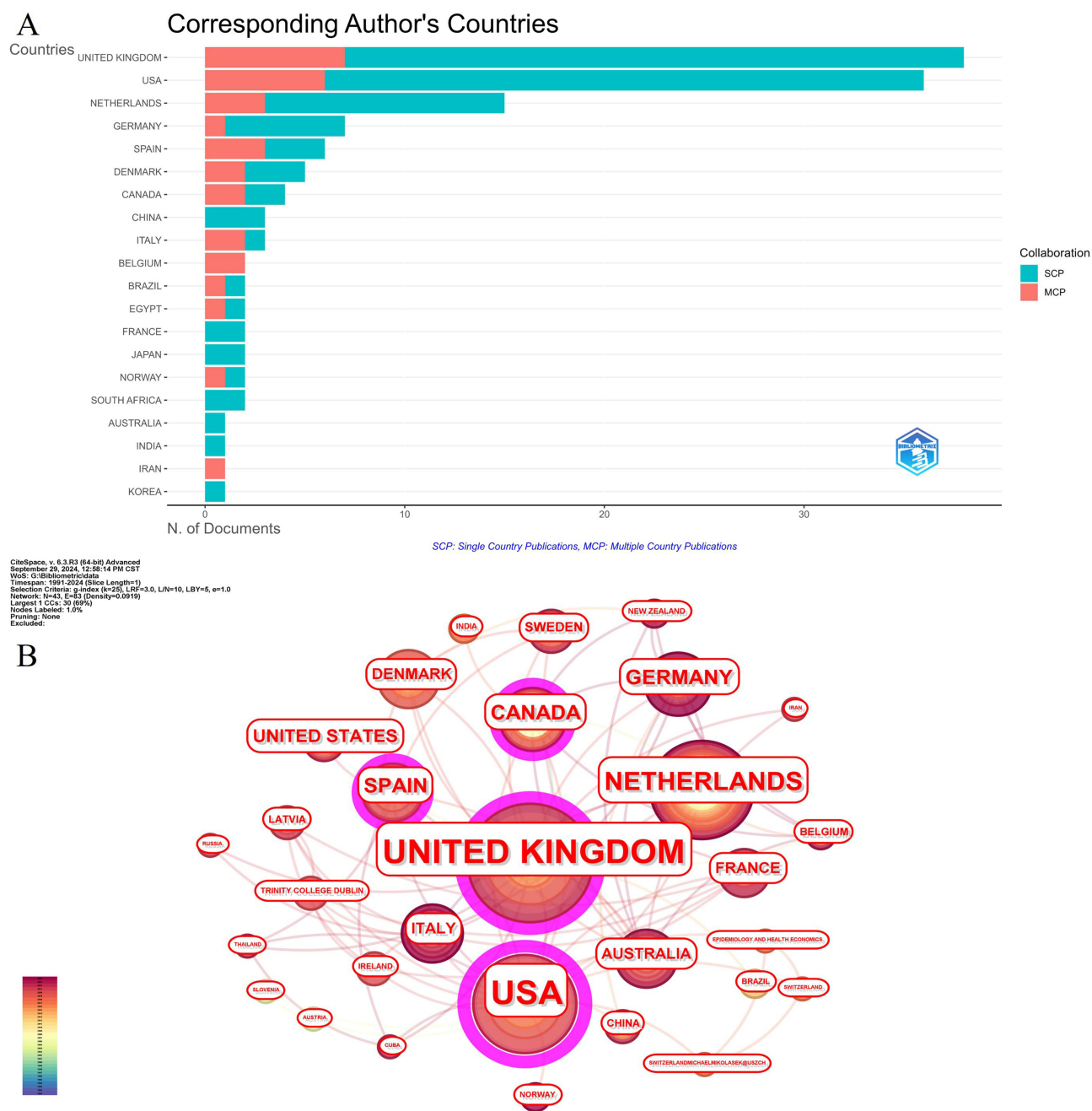**Figure 3** Interlinking of issuing journals.

## A

### Corresponding Author's Countries



CiteSpace, v. 6.3.R3 (64-bit) Advanced
September 29, 2024, 12:58:14 PM CST
WoS: G:\Bibliometric\data
Timespan: 1991-2024 (Slice Length=1)
Selection Criteria: g-index (k=25), LRF=3.0, L/N=10, LBY=5, e=1.0
Network: N=43, E=83 (Density=0.0919)
Largest 1 CCs: 30 (69%)
Nodes Labeled: 1.0%
Pruning: None
Excluded:

## B

*SCP: Single Country Publications, MCP: Multiple Country Publications*

**Figure 4** (**A**) Chart of the volume of publications by countries. (**B**) Map of cooperation networks between countries.

United Kingdom leads with the highest number of documents, indicating a significant contribution to the field. Other countries, such as the Netherlands, South Africa, Australia, and Germany, also exhibit a substantial volume of research output. The figure also emphasizes the trend of international collaboration, with an increase in the MCP suggesting a growing trend of cross-border research partnerships aimed at addressing the complexities of CFS health economics.

(Table 2) lists the top countries in terms of the frequency of publications, with England at the forefront, followed by the USA and Spain. The centrality and degree metrics in the table indicate the extent of a country's involvement and interconnectedness within the research network. The USA, with the highest centrality, is at the core of the research network, reflecting its pivotal role and extensive collaboration. The sigma value, which measures the variation in the

**Table 2** Top 10 Countries in Terms of Number of Publications

| Label | Year | Freq | Degree | Centrality | Sigma |
|---|---|---|---|---|---|
| USA | 1994 | 47 | 18 | 0.47 | 1 |
| UNITED KINGDOM | 1999 | 49 | 15 | 0.26 | 2.57 |
| SPAIN | 2010 | 10 | 9 | 0.12 | 1 |
| AUSTRALIA | 1992 | 6 | 9 | 0.03 | 1 |
| ITALY | 2020 | 6 | 9 | 0.06 | 1 |
| CANADA | 1999 | 10 | 8 | 0.11 | 1 |
| GERMANY | 2009 | 11 | 8 | 0.04 | 1 |
| BELGIUM | 2010 | 3 | 6 | 0.01 | 1 |
| FRANCE | 2004 | 5 | 6 | 0.01 | 1 |
| NETHERLANDS | 1998 | 20 | 6 | 0.02 | 1 |

number of publications, shows that while countries such as England and the USA have stable and high outputs, others exhibit variability, suggesting fluctuations in research focus or resources.

The distribution of countries/regions in the health economics evaluation of CFS underscores widespread yet uneven global engagement. Leading countries such as England and the USA are at the forefront of research, producing a substantial number of publications and driving collaborative efforts. The data highlight the importance of international cooperation, as seen in the increasing trend of the MCP, which is crucial for comprehensive and robust research in the field. The variation in publication volume and centrality among countries reflects differences in research priorities, resources, and the dynamic nature of global health economics research. This global perspective is vital for understanding the epidemiology of CFS and for developing effective health policies and economic evaluations.

## Contribution of Institutions

In the realm of health economics evaluation of chronic fatigue syndrome (CFS), various institutions have made significant contributions, shaping the discourse and advancing knowledge within the field. The data presented in Table 3 offer a glimpse into the leading organizations on the basis of their publication output, citation impact, and overall research interconnectedness.

(Table 3) highlights the top-performing institutions in terms of the number of documents published, citations received, and total link strength, which indicates the extent of their research network and collaborative ties. Aarhus University Hospital emerges as a prominent contributor, with a relatively high citation count despite a small number of publications, suggesting the impact of their work. The Adelante Centre of Expertise in Rehabilitation and Audiology and the Australian National University also stand out, with a notable total link strength indicating their active role in collaborative research efforts.

Boston University is remarkable for its substantial citation count, indicating that its publications have been widely referenced, reflecting the significance and relevance of their research. The Bristol Medical School, Population Health Sciences, with a high total link strength, has a strong presence.

**Table 3** Top 5 Organizations with Publications in the Field

| Id | Organization | Documents | Citations | Total Link Strength |
|---|---|---|---|---|
| 1 | Aarhus univ hosp | 4 | 76 | 1 |
| 6 | Adelante ctr expertise rehabil and audiol | 2 | 27 | 3 |
| 16 | Australian natl univ | 2 | 51 | 1 |
| 28 | Boston univ | 2 | 1329 | 2 |
| 31 | Bristol med sch populat hlth sci | 2 | 18 | 10 |

## Authors and Cited Authors

The analysis of leading and cited authors in the field of health economics evaluation of chronic fatigue syndrome (CFS) offers insights into the key contributors and collaborative networks within the research community. Figure 5 provides a visual representation of these dynamics, highlighting the central figures and their collaborative relationships.

(Figure 5) presents a network chart where authors are represented as nodes, and the connections between them indicate collaborations. The chart is color-coded to represent different research teams or groups, with the same color signifying collaboration. The latter figure shows that two distinct collaboration clusters are identified—one in red and one in green—suggesting the presence of two primary research teams working actively in the field.

The chart also reveals that authors such as "Hollingworth, William", "Beasant, Lucy", and "White, Peter D". are central figures with multiple connections, indicating their prolific publication records and extensive collaboration networks. These authors likely serve as thought leaders, driving the research agenda and fostering collaborative efforts. "Bleijenberg, Gijs" and "Mccrone, Ned, Paul" also appear as significant nodes, suggesting their influential roles in the field.
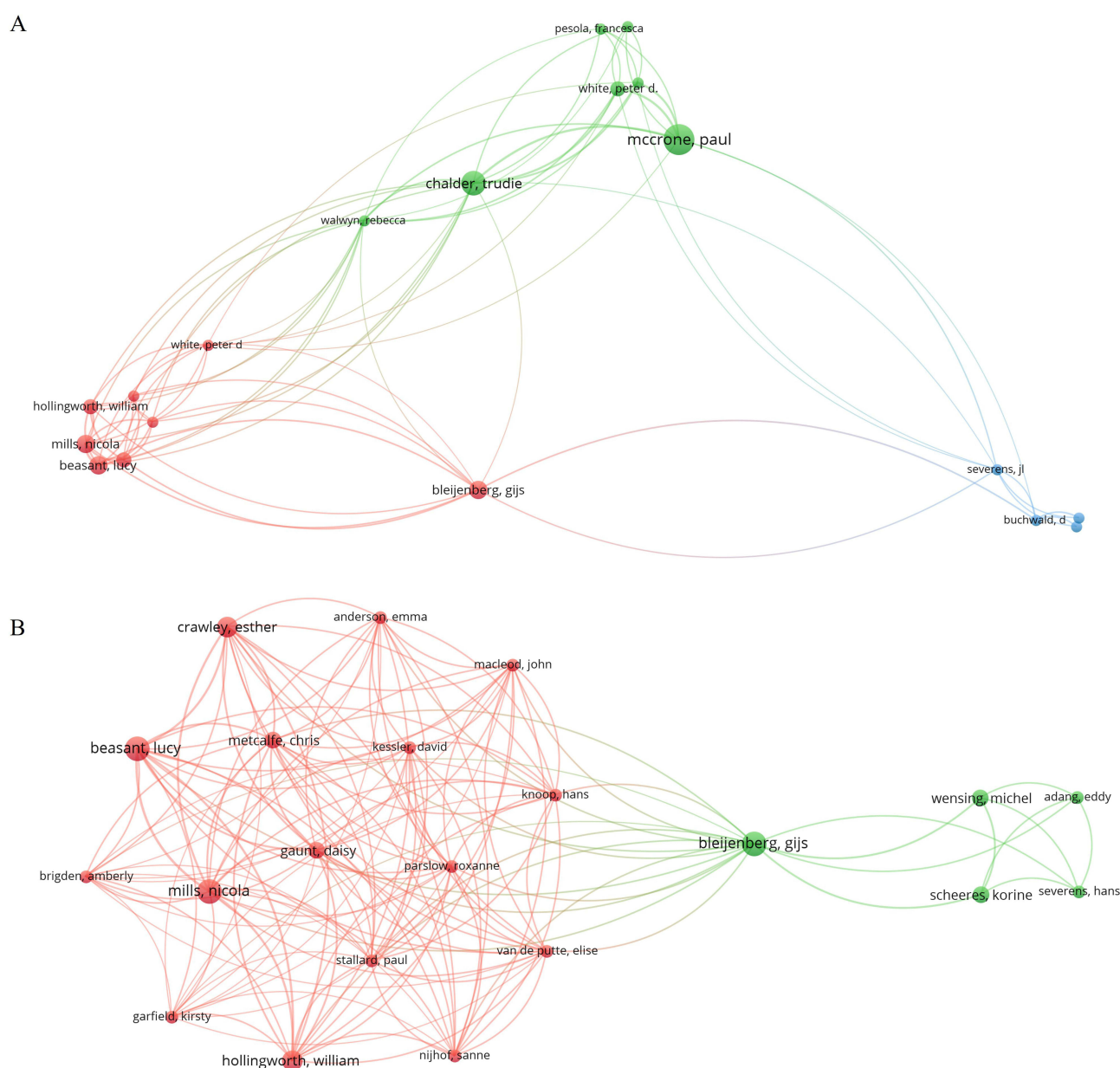


**Figure 5** (**A**) Graph of highly cited authors. (**B**) Network map of collaborating authors.

## Citation Analysis

Citation analysis is a critical aspect of understanding the influence and impact of research within the field of health economics evaluation of chronic fatigue syndrome (CFS). It provides insights into the most influential works, peer-reviewed journals, and widely cited literature in the field. Figures 6, 7, and 8 offer a detailed look into these aspects, highlighting the most cited journals, highly cited literature, and commonly cited literature, respectively.

(Figure 6) presents a visual representation of the most frequently cited journals in domains closely related to the health economics evaluation of the CFS. JAMA-J Am Med Assoc, J Psychosom Res, and New Engl J Med are prominently featured, indicating their significant influence on the field. These journals cover a broad spectrum of medical
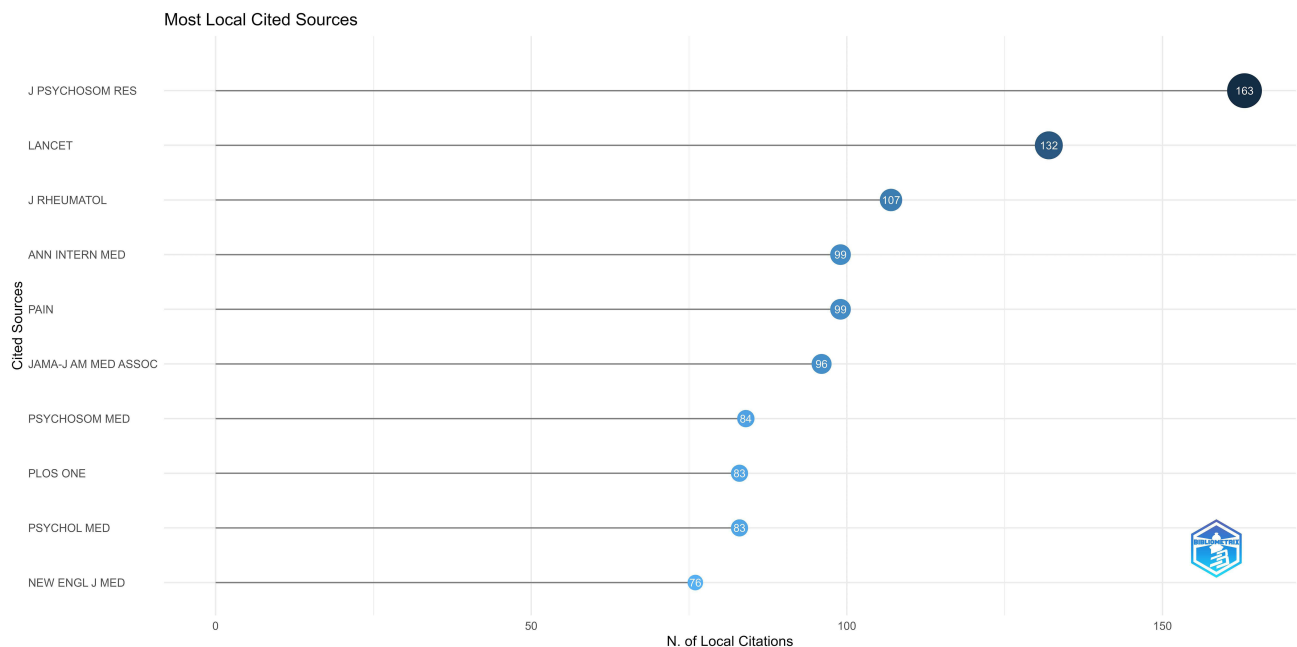


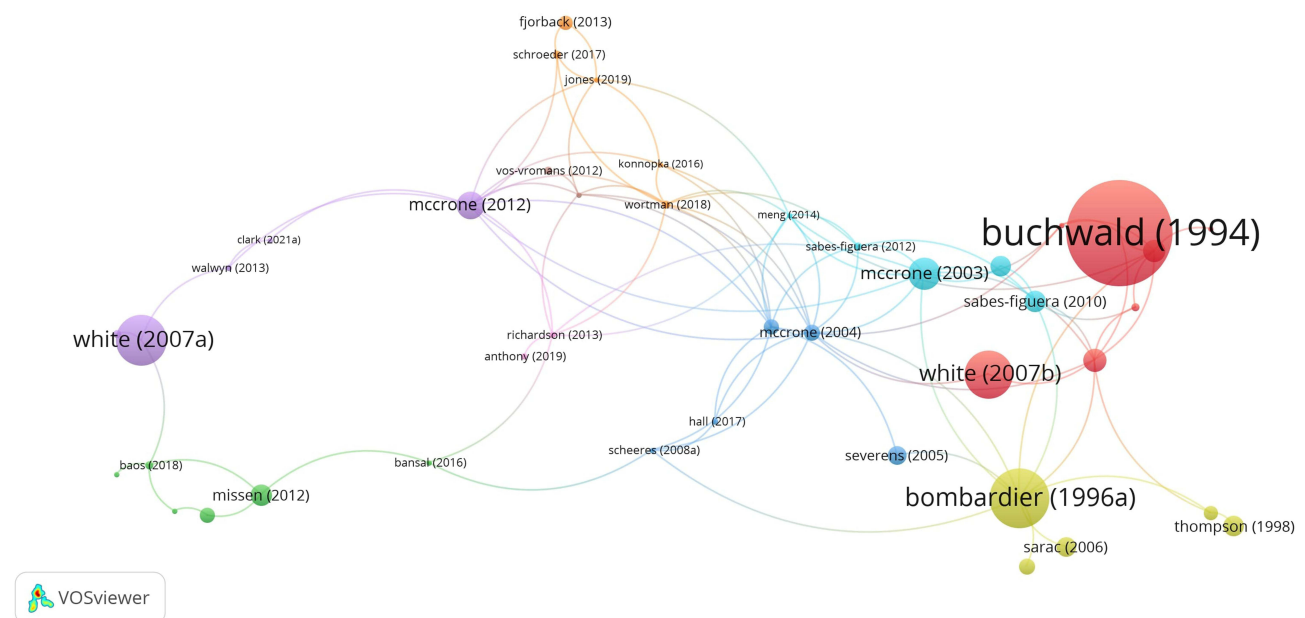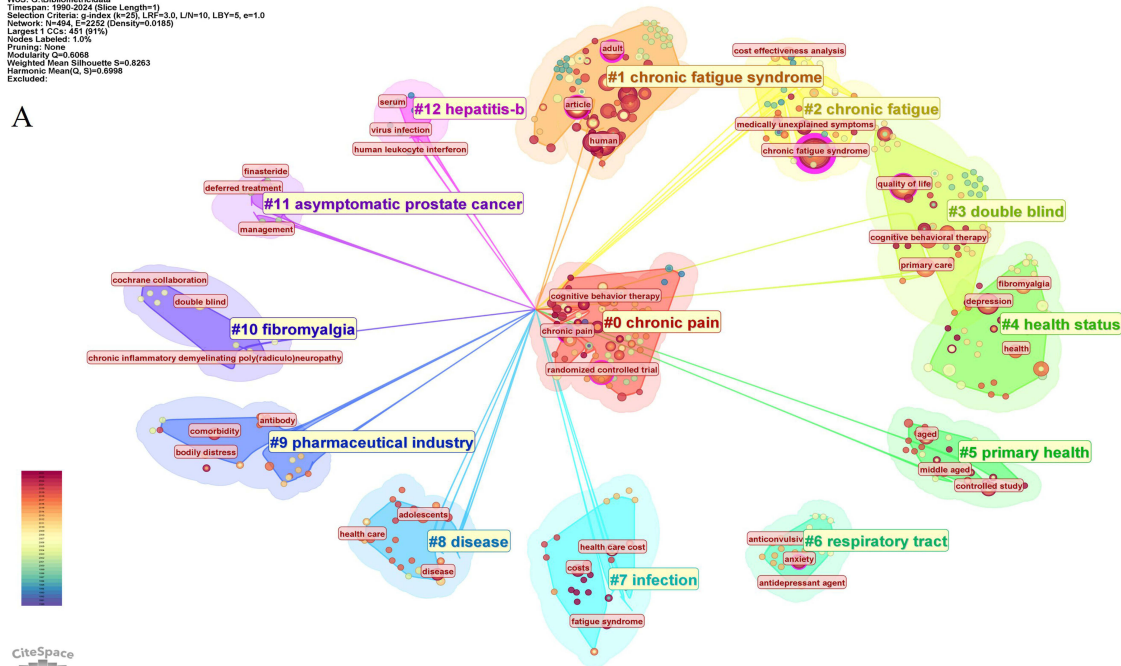**Figure 6** Most cited journals in neighboring domains.



**Figure 7** Highly Cited Literature Chart.

**Figure 8** Commonly Cited Literature Chart.

and health-related topics, and their high citation counts reflect the depth and breadth of the research they publish. The figure also includes other notable journals, such as Psychol Med, PLOS ONE, and Psychosom Med, which are recognized for their contributions to the discourse on CFS and related health economic issues.

(Figure 7) focuses on individual articles or papers that have been highly cited within the field. This chart highlights the most impactful studies, as measured by the number of citations they have received. The presence of works by authors such as White (2007a), Bao (2018), and Walwyn (2013) on this chart underscores the importance of their research in shaping the understanding of CFS health economics. These highly cited papers likely address key issues, present novel methodologies, or offer significant insights that have been valuable to other researchers in the field.

(Figure 8) provides an overview of the literature that is commonly cited across various studies in the field. This chart includes works by authors such as Goldberg (2002), Briggs (1997), and McCrone (2003, 2004), indicating their enduring relevance in the field. The inclusion of these works suggests that they provide foundational knowledge, established methodologies, or comprehensive reviews that are frequently referenced by researchers. The commonly cited literature serves as a backbone for the field, offering a solid base of knowledge upon which current and future research is built.

## Keyword Analysis

Keyword analysis is pivotal for identifying the focal points and intellectual progress within research on health economics evaluations of chronic fatigue syndrome (CFS). Figures 9 and 10, which consist of multiple graphs, offer a visual synthesis of the thematic trends and scholarly attention over time.

The keyword clustering graph in (Figure 9) illustrates the thematic groupings of keywords that frequently appear together in the CFS health economics literature. These clusters represent the main research themes and their inter-connections, providing a snapshot of the research areas that are closely related. The timeline graph within Figure 9 complements this by showing the evolution of these themes over time, highlighting periods of increased interest or significant shifts in research focus.

(Figure 10) presents an explosion chart that tracks the emergence and prominence of specific keywords within the CFS literature. This chart is instrumental in identifying sudden surges in citations for particular terms, indicating
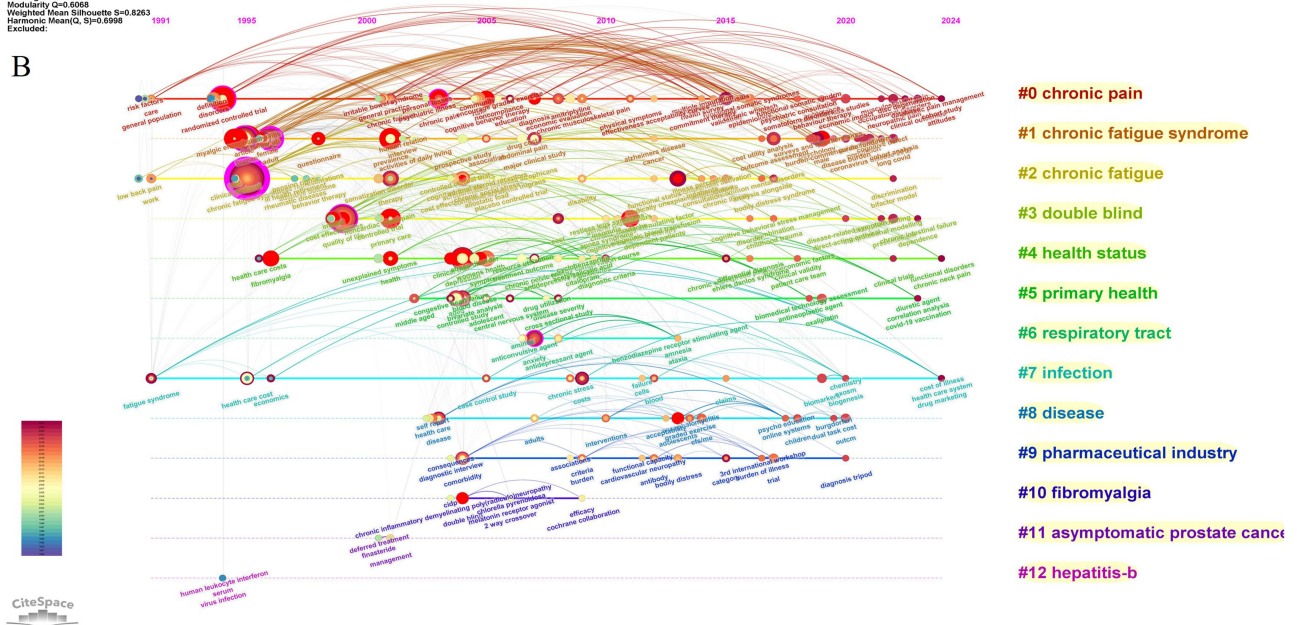
Figure 9 (A) Keyword clustering map. (B) Timeline graph of keywords highlighted in the clustering.

a "burst" of scholarly interest. The chart reveals keywords such as "cost effectiveness analysis", "clinical trial", and "fibromyalgia" experiencing significant bursts, suggesting that these areas have been intensively studied during certain periods.
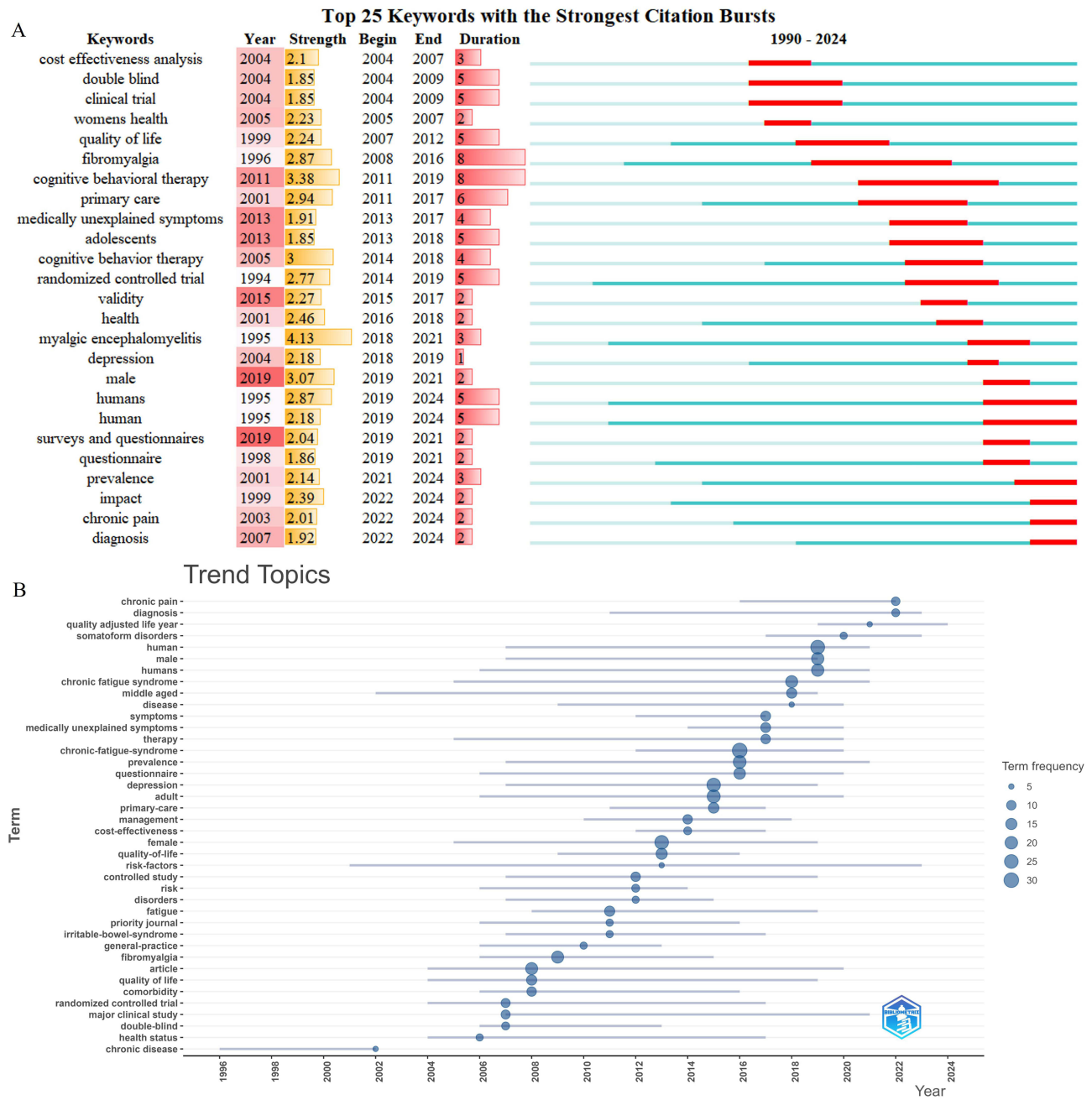
## Figure 10

**A**

### Top 25 Keywords with the Strongest Citation Bursts

| Keywords | Year | Strength | Begin | End | Duration | 1990 - 2024 |
|---|---|---|---|---|---|---|
| cost effectiveness analysis | 2004 | 2.1 | 2004 | 2007 | 3 | |
| double blind | 2004 | 1.85 | 2004 | 2009 | 5 | |
| clinical trial | 2004 | 1.85 | 2004 | 2009 | 5 | |
| womens health | 2005 | 2.23 | 2005 | 2007 | 2 | |
| quality of life | 1999 | 2.24 | 2007 | 2012 | 5 | |
| fibromyalgia | 1996 | 2.87 | 2008 | 2016 | 8 | |
| cognitive behavioral therapy | 2011 | 3.38 | 2011 | 2019 | 8 | |
| primary care | 2001 | 2.94 | 2011 | 2017 | 6 | |
| medically unexplained symptoms | 2013 | 1.91 | 2013 | 2017 | 4 | |
| adolescents | 2013 | 1.85 | 2013 | 2018 | 5 | |
| cognitive behavior therapy | 2005 | 3 | 2014 | 2018 | 4 | |
| randomized controlled trial | 1994 | 2.77 | 2014 | 2019 | 5 | |
| validity | 2015 | 2.27 | 2015 | 2017 | 2 | |
| health | 2001 | 2.46 | 2016 | 2018 | 2 | |
| myalgic encephalomyelitis | 1995 | 4.13 | 2018 | 2021 | 3 | |
| depression | 2004 | 2.18 | 2018 | 2019 | 1 | |
| male | 2019 | 3.07 | 2019 | 2021 | 2 | |
| humans | 1995 | 2.87 | 2019 | 2024 | 5 | |
| human | 1995 | 2.18 | 2019 | 2024 | 5 | |
| surveys and questionnaires | 2019 | 2.04 | 2019 | 2021 | 2 | |
| questionnaire | 1998 | 1.86 | 2019 | 2021 | 2 | |
| prevalence | 2001 | 2.14 | 2021 | 2024 | 3 | |
| impact | 1999 | 2.39 | 2022 | 2024 | 2 | |
| chronic pain | 2003 | 2.01 | 2022 | 2024 | 2 | |
| diagnosis | 2007 | 1.92 | 2022 | 2024 | 2 | |

**B**

### Trend Topics



**Figure 10** (**A**)Top 25 Keywords with the Strongest Citation Bursts. (**B**) Timeline chart of highlighted Trend Topics.

## Discussion

### General Information

The bibliometric analysis presented in this study offers a comprehensive overview of the trends and hotspots in the health economics evaluation of chronic fatigue syndrome (CFS). Our research has systematically cataloged the burgeoning body of literature on CFS, revealing significant growth in scholarly output over the past three decades. The temporal distribution graph of the literature indicates an upward trajectory in publications, with a notable surge in research activity from the 2000s onward, suggesting an increase in research interest and awareness of the health economic impact of CFS.

The analysis of the global publication trend reveals a rich tapestry of influential journals contributing to the discourse on CFS. As depicted in Figure 6, journals such as JAMA-J Am Med Assoc, J Psychosom Res, and New England J Med

stand out for their high citation counts, underscoring their pivotal roles in shaping the field. These platforms have been instrumental in disseminating influential studies that have propelled forward the understanding and evaluation of the health economics of CFS. Table 1, which shows the top influential journals in the field, highlights the substantial contribution of journals such as BMC Health Services Research and BMC Neurology. These journals have not only published a significant number of documents but also garnered considerable citations, reflecting their substantial impact on the CFS research community. The high citation count for Clinical Infectious Diseases, despite a low total link strength, may indicate the presence of highly influential yet independently cited works within the CFS health economics discourse. Figures 7 and 8 further refine our understanding of the research landscape by highlighting the most and most commonly cited literature, respectively. These figures reveal a corpus of foundational and impactful works that have significantly influenced the trajectory of CFS research. The high citation bursts for certain keywords, as shown in Figure 10, indicate focal areas of intense scholarly interest and potential research breakthroughs within the field of CFS health economics.

## Keywords and Trends: Insights from the Data

In health economics research, common research methods include cost-minimization analysis (CMA), cost-effectiveness analysis (CEA), cost-utility analysis (CUA), and cost–benefit analysis (CBA.[17–20] This study revealed that CEA is currently the main method used in health economics research on CFS. The CEA evaluates the economic efficiency of interventions by calculating the cost per unit of health gain (eg, additional years of life per year, avoided days of illness per day, etc).[21] CEA can help determine which treatment options can result in the greatest health gains when resources are limited. For example, cognitive behavioral therapy (CBT) has been shown to be a relatively more cost-effective intervention for people with CFS, especially when it is measured in terms of quality-adjusted life years (QALYs). CBT is relatively cost-effective and can effectively optimize the allocation of health resources for people with CFS.[22] CEA not only considers medical costs but also incorporates the loss of patient productivity and the cost of informal care into the calculation, thereby providing a comprehensive economic evaluation. This is important for assessing the impact of chronic fatigue syndrome on patients' quality of life and the overall social cost of different treatments.[23] In addition, CEA can be used to compare multiple treatment options, such as cognitive behavioral therapy, to help decision-makers choose the optimal treatment that can both reduce symptoms and save costs.[24] In summary, CEA can be used in the future to evaluate the health economics of CFS patients, which can help optimize the use of medical resources while ensuring that patients achieve the best possible health outcomes.

According to the keyword prominence map, chronic fatigue syndrome and depression are both highly expressed keywords, suggesting that there is a certain connection between chronic fatigue syndrome and depression at the level of health economics. Studies have shown that up to two-thirds of chronic fatigue syndrome patients also suffer from depression or anxiety. These patients often require frequent medical visits, drug treatment, and long-term psychological health management, which significantly increase their medical costs. The impact of this comorbidity on work productivity and quality of life further increases the socioeconomic burden.[25] The role of depression in chronic fatigue syndrome is not limited to mood disorders. It is also closely related to the patient's functional impairment. Depression may significantly increase the risk of disability in CFS patients by exacerbating fatigue, which in turn increases the demand for medical resources and increases the cost of hospitalization, rehabilitation and psychotherapy, further increasing the economic burden on patients.[26] In addition, the coexistence of chronic fatigue syndrome and depression can lead to serious social dysfunction, especially a decline in social interaction and occupational productivity, which in turn leads to long-term unemployment or reduced work efficiency, further increasing the economic burden.[27] Therefore, from the perspective of health economics, when treating patients with chronic fatigue syndrome, attention should also be given to whether they have other forms of psychological distress. In particular, early screening for depressive symptoms can be carried out for early detection, intervention and treatment, with the aim of addressing the patient's pain while reducing the economic burden on the patient, their family and society.

In the treatment of chronic fatigue syndrome, keyword highlighting suggests that there have been more studies on the health economic evaluation of cognitive behavioral therapy and primary care. On the one hand, future research could be conducted on the health economic indicators of other treatments for chronic fatigue syndrome to determine whether other

treatment methods have an advantage in treating chronic fatigue syndrome at the level of health economics. On the other hand, these findings also suggest that cognitive behavioral therapy and primary care may indeed have certain advantages in terms of health economics when treating chronic fatigue syndrome. Studies have shown that CBT has significant clinical effects in treating CFS patients, can reduce fatigue symptoms and improve patients' daily functions. Compared with other psychological treatments, CBT, in particular, has greater treatment effects in terms of reducing the use of medical resources and improving patients' quality of life and can effectively reduce medical costs.[28] CBT also has a significant effect on long-term fatigue management and quality of life improvement in CFS patients, thereby reducing their long-term dependence on medical resources and further reducing social costs.[29] In addition, patients treated with CBT have significant improvements in social functioning and occupational productivity, which can effectively reduce indirect costs from a health economics perspective, including expenditures on unemployment insurance and lost productivity.[30] Therefore, CBT not only effectively alleviates patients' symptoms when treating CFS but also has definite advantages in terms of health economics by reducing the use of medical resources and improving cost-effectiveness. According to existing studies, primary care has certain advantages in terms of health economics when managing chronic fatigue syndrome, as it can control costs, improve patients' quality of life and work ability, and thus reduce long-term social and economic burdens. For example, early intervention by primary care can effectively prevent the aggravation of CFS symptoms, thereby reducing the need for later specialist treatment and high-cost medical services such as hospitalization. Studies have shown that early intervention in primary care, such as psychoeducation and cognitive behavioral therapy (CBT), can effectively control the condition before symptoms worsen, thereby reducing long-term medical expenses.[31] Treatment through primary care can also ensure more continuous and convenient care for patients, especially through telemedicine and regular follow-up. Primary care providers can establish long-term therapeutic relationships with patients, which can help improve treatment compliance and ensure that patients continue to receive effective treatment while avoiding the complexity and waiting times of specialist care.[32] In addition, primary care often involves multidisciplinary interventions, including nutrition, exercise and psychological support, which can comprehensively improve patients' quality of life and significantly reduce the subsequent high-cost consumption of medical resources. For example, a primary care programme that combines nutrition, chronobiology, and exercise interventions not only improved fatigue and pain symptoms in CFS patients but also reduced healthcare expenditures and indirect costs such as unemployment or reduced work capacity.[33] In summary, primary care not only provides efficient cost control when managing chronic fatigue syndrome but also improves patients' quality of life and work capacity, reducing long-term social and economic burdens.

## Limitations

This paper has the following limitations. Bibliometrics is a method of analyzing the literature on the basis of published literature that can be analyzed and mined for potential directions for analysis. This paper incorporates three major databases commonly searched, WOSCC, PubMed, and Scopus, but does not include all the databases, which may have resulted in the omission of certain topics in this field category. Moreover, the data from the WOSCC, PubMed and Scopus databases are not uniform. The PubMed database is not a citation database, and there are no "cocited literature" data, so it is not capable of "cocitation analysis" of the literature.

## Conclusion

This study highlights the use of health economics in chronic fatigue syndrome through a bibliometric analysis, highlighting this approach as an important method for future research and evaluation. Moreover, in previous studies on chronic fatigue syndrome, although the mechanism of its etiology has been difficult to elucidate systematically, we can intervene in terms of outcomes through a health economics approach to mitigate the harm caused by chronic fatigue syndrome. In addition, as we move forward, it is important not only to deepen our scientific inquiry but also to promote cross-border collaboration to accelerate the development of effective strategies to systematically understand chronic fatigue syndrome and reduce its global burden.

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