Quality of life in patients with Sjögren's disease: a bibliometric analysis

W. Liu, Z. Guo, X. Wang, N. Lu, X. Tang, Q. Jiang, X. Zhou

Department of Rheumatology, Guang'anmen Hospital, China Academy of Chinese Medical Sciences, Beijing, China.

Abstract

Objective

Quality of life (QoL) in patients with Sjögren's disease (SjD) is a critical area of research that demands attention due to the impact of SjD on patients' lives. This study utilised bibliometric methods, aiming to comprehensively explore the research content and hotspots in the field of QoL in patients with SjD.

Methods

The literature data source for this study was the Web of Science Core Collection. CiteSpace and VOSviewer were used to analyse publications in relation to authors, countries, institutions, journals, references, and keywords.

Results

The study focused on literature that addressed QoL in SjD patients, involving a total of 922 authors and 336 articles published across 151 journals. The study revealed that the number of publications in this field has remained relatively low, exhibiting a stable yet gradual upward trend, with no evidence of explosive growth. Key journals in this field include the Journal of Annals of the Rheumatic Diseases, Rheumatology (Oxford), Clinical and Experimental Rheumatology, and the Journal of Arthritis & Rheumatology. Asghar Bowman Simon J is the most prolific author in the field (21 publications), and England and the University Hospitals Birmingham NHS Foundation Trust and University of Groningen have the most publications. The most high-frequency keywords are "Sjögren's syndrome", "quality of life", "fatigue", "xerostomia", "depression", "sexual dysfunction" and "xerophthalmia".

Conclusion

This study represents a bibliometric analysis focusing on QoL in patients with SjD. It underscores the need for more extensive and systematic research in this area, emphasising the importance of a multidisciplinary approach. Despite advancements in medical research for SjD, there is a crucial need to focus on QoL to enhance patient satisfaction and overall well-being. The findings advocate for more personalised treatment plans and a better understanding of the psychosocial needs of patients with SjD to improve their quality of life.

Key words

Sjögren's syndrome, quality of life, bibliometric analysis, citespace, vosviewer

Wenjing Liu, MM
Zilin Guo, MD
Xuanyun Wang, MM
Nannan Lu, MM
Xiaopo Tang, MD
Quan Jiang, MD
Xinyao Zhou, MD

Please address correspondence to:
Xinyao Zhou
Department of Rheumatology,
Guang'anmen Hospital,
China Academy of
Chinese Medical Sciences,
100053 Beijing, China.
E-mail: xyz_1102@126.com

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Introduction

Sjögren's disease (SjD) is a systemic autoimmune disease characterised by chronic inflammation of the salivary and lachrymal glands that causes dryness of the eyes and of the mouth. Besides dryness, the other two key symptoms of the disease are fatigue and widespread pain. In addition, extraglandular manifestations are present in 30–40% of patients, such as interstitial lung disease, peripheral neuropathy, and interstitial nephritis (1-5). SjD is a heterogeneous and disabling condition that negatively affects the quality of life (QoL) of the patient and increases both morbidity and mortality (3, 4, 6, 7). Although most patients experience poor QoL due to symptoms such as fatigue and pain, the use of traditional disease-modifying antirheumatic drugs (DMARDs) is primarily based on experience from other connective tissue diseases and therefore is not truly suitable for patients with SjD (8). A burgeoning effort is underway to subgroup SjD using endotypic biomarkers to stratify patients by pathophysiology, autoantibodies specificity, and cytokine and/or molecular profiling, going beyond the traditional phenotypic classification (9). Identifying specific pathogenic targets will enable physicians to achieve personalised treatment, thereby better controlling symptoms, improving patients' QoL, and reducing the risk of permanent organ damage (10-15). Significant efforts are currently underway internationally to design clinical trials that take into account the heterogeneity of patient populations and the potential discrepancies between systemic disease activity and patient-reported outcomes (15, 16). Importantly, new outcome measurement tools have been proposed to address the challenges encountered in defining patient treatment responses (17).

Quality of life refers to an individual's sense of well-being, life purpose, autonomy, the ability to fulfil meaningful roles, and the ability to engage in important relationships (18). It encompasses physical health, material health, social health, emotional health, development and activities (19). DeRidder *et al*. mentioned in the process of psychological adaptation to chronic diseases that chronic illnesses cause significant changes in a patient's life, which negatively affect their quality of life and well-being (20). Commonly used scales include the EuroQol 5-Dimensional Health Scale (EQ-5D) (21), the Short Form Health Survey SF-36 (22), the Hamilton Anxiety Scale (HAMA) (23), and the Hamilton Depression Scale (HAMD) (23). Functional status, depression, social support, and age are significant predictors of quality of life, with functional status and depression being the most critical determinants (24). SjD patients, regardless of whether they are in an active disease phase, experience a lower quality of life compared to the general population. The primary factors affecting the quality of life in SjD patients are various clinical symptoms. Due to reduced function of the lacrimal and salivary glands, patients experience discomfort such as itching, photophobia, and difficulty with daily activities like reading and driving, as well as the subjective sensation caused by a lack of saliva in the mouth. These are the main reasons for the decline in quality of life in SjD patients (25-28). Studies have also found that delayed medical visits and diagnostic delays worsen the condition, further reducing quality of life (29-32). Other symptoms caused by dryness, such as dysphagia (33, 34), taste dysfunction (35, 36), voice disorders (37, 38), sexual dysfunction (39-41), and skin itching (42, 43), also negatively impact quality of life. Systemic symptoms such as pain (44, 45), fatigue (46, 47), sleep disorders (48, 49), as well as psychological symptoms like anxiety/depression (50-52) and feelings of loneliness (53), make the quality of life of SjD patients significantly lower than that of the general population. Research has shown that SjD patients with anxiety/depression tend to adopt negative coping strategies to solve problems in both work and life, leading to limitations in their work and daily life, and a further decline in quality of life (51).

However, despite some progress in relevant research, a comprehensive analysis of authors, institutions, countries, and research trends in the field of QoL

studies on SjD patients remains insufficient. Bibliometrics, as a commonly used method, can analyse the associations between authors and institutions, revealing global research progress and trends (54). This approach helps researchers quickly identify current hotspots in the field, integrate research information, and promote communication and collaboration among researchers. Therefore, we will adopt bibliometric methods to integrate the current research on QoL in the field of SjD, with the aim of attracting more attention from researchers, doctors, and caregivers to the healthy lives of SjD patients and deepening the understanding of their health conditions. This will enable us to provide customised and targeted interventions, as well as comprehensive care models, including psychosocial support, counselling, and educational programs, thereby comprehensively improving the QoL of SjD patients.

To the best of our knowledge, this is the first bibliometric study based on the Web of Science Core Collection (WoSCC) that evaluates the QoL of patients with SjD using bibliometric and visualisation analysis methods. To verify this, we performed a systematic literature search in the Web of Science database using the following search strategy: [TS=("sicca syndrome") OR TS=("Sjogren's syndrome") OR TS=("Sjögren's Syndrome") OR TS=("Sjögren Syndrome") OR TS=("Sjögrens Syndrome") OR TS=("Sjögren's disease")]AND[TS=("Quality of life") OR TS=("Health-Related Quality Of Life") OR TS=("HRQoL") OR TS=("Life Quality") OR TS=("QoL") OR TS=("well-being") OR TS=("SF-36") OR TS=("EQ-5D")]AND[TS=("bibliometric") OR TS=("science mapping") OR TS=("visualization analysis") OR TS=("co-word analysis") OR TS=("co-occurrence analysis") OR TS=("co-citation analysis") OR TS=("VOS viewer") OR TS=("CiteSpace") OR TS=("biblioshiny")],

No prior bibliometric review addressing this specific topic was identified, highlighting both the necessity and the novelty of the present study.

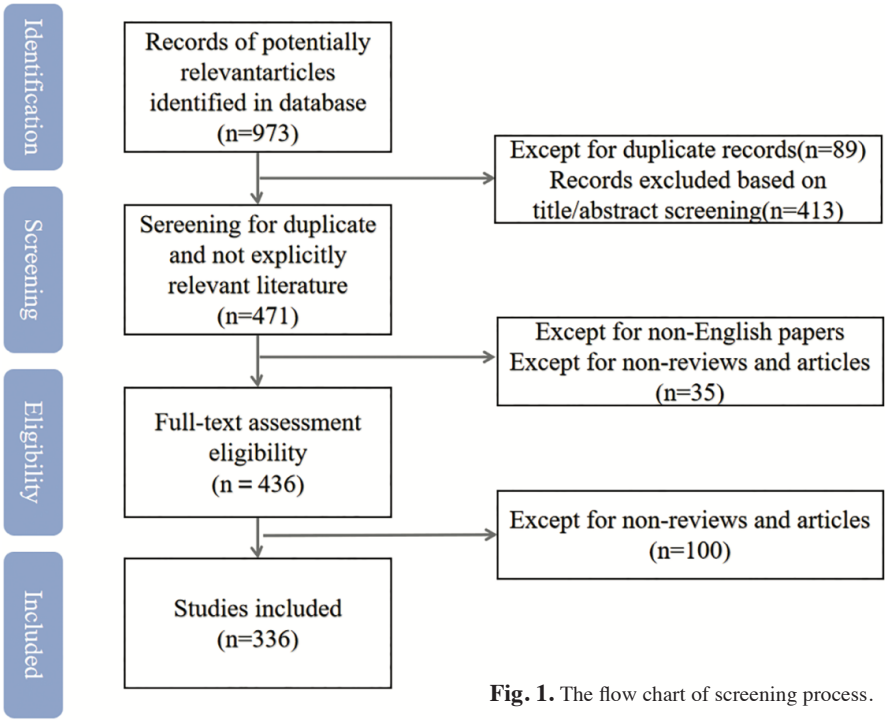

Fig. 1. The flow chart of screening process.

Methods

Literature retrieval and data collection

The Web of Science Core Collection (WoSCC) was used as the source of bibliographic data. Web of Science has been widely accepted by researchers as a high-quality digital literature resource database and is generally considered the most suitable database for bibliometric analysis, and the citation index was chosen as Science Citation Index Expanded (SCI-EXPANDED) -1900-present in order to ensure comprehensive and accurate search data. Search strategy: (TS=("sicca syndrome") OR TS=("Sjogren's syndrome") OR TS=("Sjögren's Syndrome") OR TS=("Sjögren Syndrome") OR TS=("Sjögrens Syndrome") OR TS=(" Sjögren's disease"))AND (TS=("Quality of life") OR TS=("Health-Related Quality Of Life") OR TS=("QoL") OR TS=("HRQoL") OR TS=("Life Quality") OR TS=("QoL") OR TS=("well-being")OR TS=("SF-36")OR TS=("EQ-5D")).

The search period was limited to January 1, 2004, to December 31, 2024, with article and review selected as the document type and English as the language.

Data analysis

We used VOS Viewer 1.6.20 and Microsoft Excel 2021 for data deduplication, integration, and visual analysis. CiteSpace 6.1.R1 and VOS Viewer 1.6.20 were used to produce network graphs to extract and analyse the number of publications (including output, authors, journals, countries, and institutions), citation frequency (including co-cited authors, and co-occurring keywords to track research trends and hotspots. Each software is information visualisation software based on the Java platform. In the presented visual graphs, each node represents a different parameter, including countries, institutions, authors, and keywords. The weight of a parameter determines the size of its node, the heavier the weight, the larger the node. Nodes and lines are assigned different colours based on their clusters or timelines. The distance between any two nodes indicates their correlation, and the thickness of the connecting line represents the strength of the link. We also used SCImago Graphica Beta 1.0.46 to create a world map showing the distribution of countries or regions involved in research in this field.

Research ethics

The study was conducted as a biblio-

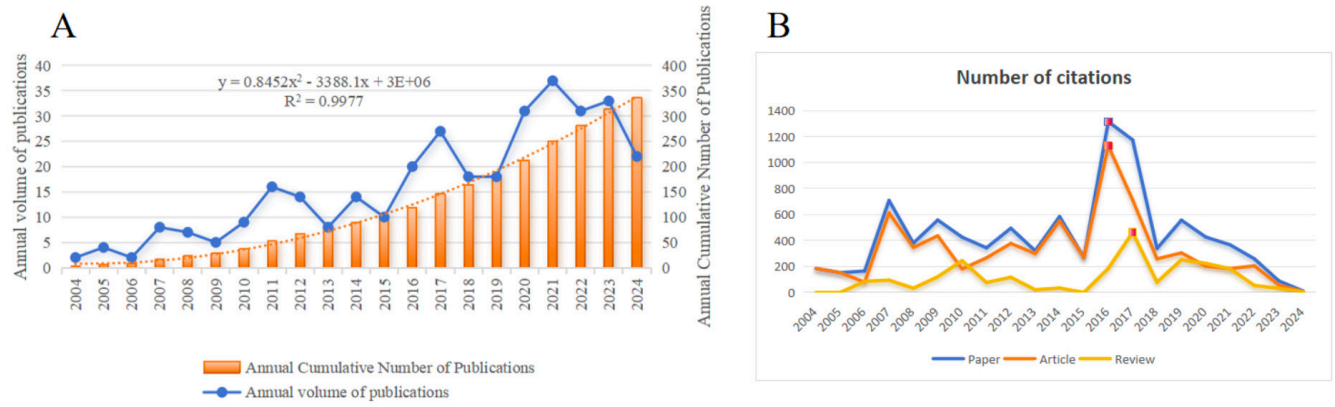

Fig. 2. The number of publications and citations.
A: The number of publication outputs and growth trend from 2004 to 2024. **B**. The number of annual citations from 2004 to 2024.

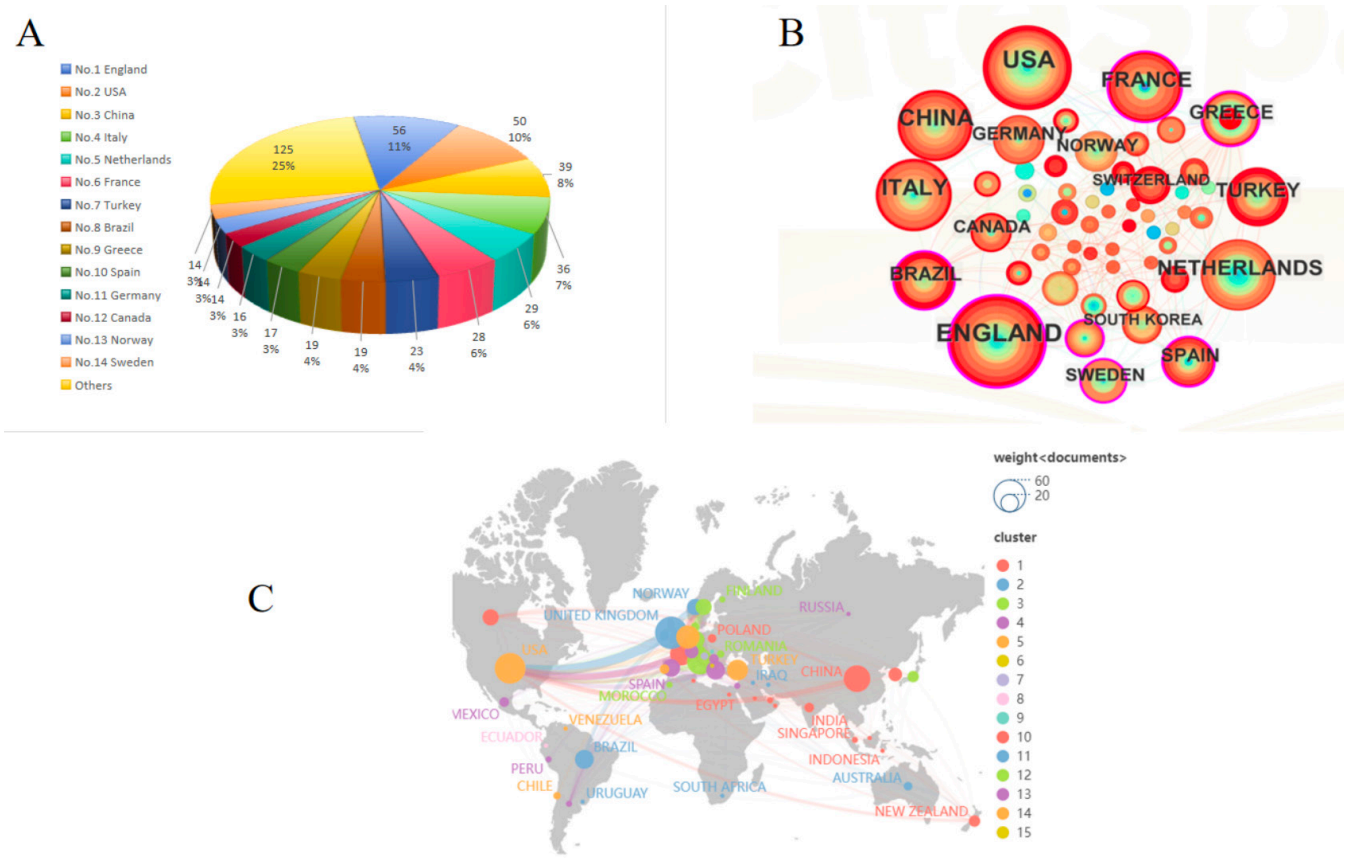

Fig. 3. The number of countries. **A**: The top 14 productive countries with publications concerning QoL in SjD. **B**: The co-occurrence map of countries concerning QoL in SjD. **C**: The distribution of countries/region.

metric analysis. All data sources were available on the Internet, and no animal or human subjects were involved. Therefore, permission was not required from the ethics committee.

Results

Analysis of publication outputs and citations

Between January 1, 2004, and December 31, 2024, the Web of Science Core Collection included 336 articles that met the inclusion criteria, including 259 articles and 77 reviews, with an average annual yield of 17 articles (Fig. 1). We obtained graphs of the number of publications and citations (Fig. 2) using Microsoft Excel. Although the annual publication output has fluctuated, the polynomial fit of the cumulative number of publications demonstrated a highly consistent linear growth pattern (R^2=0.9977, Fig. 2A), indicating that research activity in this field has progressed at a stable and predictable pace over the past two decades. Figure 2B shows that 336 papers were cited 9130 times (H-index 52), with an average citation per paper of 27.17 and an average annual citation frequency of 435, with 1316 citations in 2016, the highest ever, and the citation volume of 'Articles' dominates in most years, indicating the primary contribu-

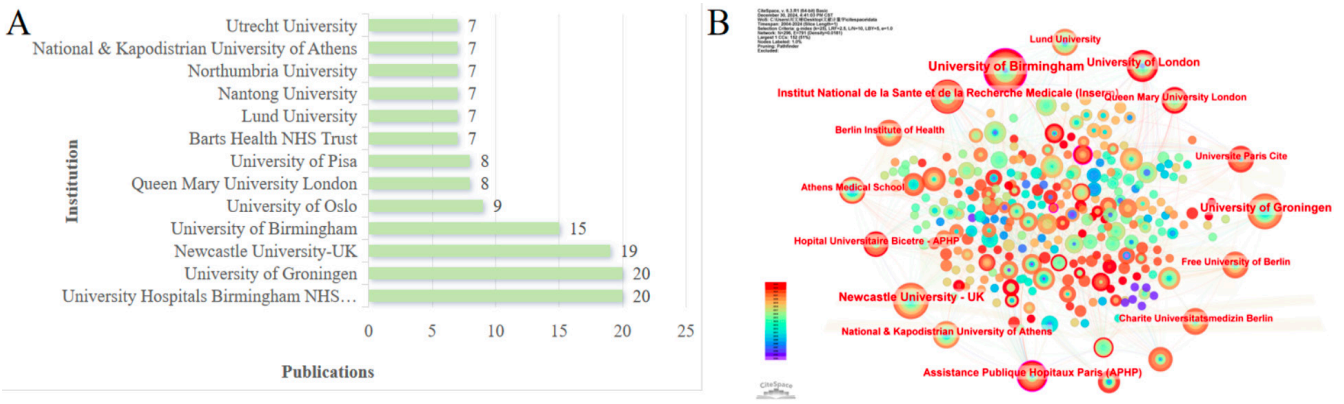

Fig. 4. The number of institutions. **A**: The top 11 productive institutions with publications concerning QoL in SjD. **B**: The analysis of research institutions concerning QoL in SjD.

tion of original research to citations. Overall, the current research on Quality of life in patients with SjD is limited in volume and lacks an explosive growth trend, indicating the need for more prospective research in this field.

Analysis of countries and institutions

An analysis of the country collaboration network through CiteSpace and Microsoft Excel (Fig. 3A-B) showed that the research on quality of life in patients with SjD involved 56 countries/regions. The England led the field with 56 published articles, followed by the USA with 50 articles. Other countries with significant contributions include China (n=39), Italy (n=36), Netherlands (n=29), France (n=28), Turkey (n=23), Brazil (n=19), Greece (n=19), Spain (n=17), Germany (n=16), Canada (n=14), Norway (n=14), and the Sweden (n=14), with a collective contribution of 225 articles from other countries. The top five countries in terms of publication quantity, excluding China, which is a developing country, are all developed countries. This indicates that developed countries have played a significant role in this research field. The purple circle represents centrality, and centrality greater than 0.1 is considered an important node. The England (0.29), Greece (0.28), Spain (0.20), Netherlands (0.14) and France (0.12) are the top six countries in terms of centrality and have an important impact on global scientific research cooperation. The countries or regions contributing in this area are shown in the world map in Figure 3C.

The top ten institutions in terms of publication volume are the University Hospitals Birmingham NHS Foundation Trust (n=20), University of Groningen (n=20), Newcastle University-UK (n=19), University of Birmingham (n=15), University of Oslo (n=9), Queen Mary University London (n=8), University of Pisa (n=8), Barts Health NHS Trust (n=7), Lund University (n=7), Nantong University (n=7), Northumbria University (n=7), National & Kapodistrian University of Athens (n=7) and Utrecht University (n=7) (see Fig. 4A for details). The centrality of the University of Birmingham exceeds 0.1, establishing a wide and close collaborative relationship with other institutions (see Fig. 4B for details).

Analysis of authors and co-cited authors

A total of 922 authors have contributed to the research on quality of life in SjD. From the perspective of authors, Bowman from University Hospitals Birmingham NHS Foundation Trust have the highest publication volume, with a total of 21 related articles. They have been involved in assessing the quality of life of SjD patients and are dedicated to improving treatment regimens to alleviate symptoms such as dry mouth and dry eyes, thereby enhancing patients' quality of life (17, 55-57). They also focus on the impact of fatigue on the quality of life of SjD patients (58). Together with Ng, the third most prolific author, they discussed the impact of dry mouth and fatigue symptoms on SjD patients' quality of life. Ng primarily researches the mechanisms of fatigue in SjD patients and explores its biological basis (59, 60). Seror from Paris-Saclay University has the highest citation count. Her research has validated SjD disease activity assessment tools such as the EULAR Primary Sjögren's Syndrome Disease Activity Index (ESSDAI) and the Patient Reported Index (ESSPRI), as well as the Sjögren's Syndrome Assessment Tool (STAR), providing reliable tools for evaluating the quality of life of SjD patients. Her work also highlights the impact of psychosocial factors, such as anxiety and depression, on the quality of life of SjD patients in clinical assessments (17, 55). Additionally, she emphasises the importance of personalised and targeted therapies in the management of SjD, particularly in alleviating dry symptoms and improving quality of life, such as the use of artificial tears and oral moisturisers (61). These studies have greatly promoted academic attention to the quality of life of SjD patients, as well as more scientific management and care for these patients. Overall, the close collaboration among highly-output and highly-cited authors has accelerated the rapid development of research in this field (Table I).

Figure 5 illustrates the collaborative relationships among authors. It shows a close collaboration between Bowman, Ng and Bombardieri, as well as between Baldini and Bootsma having a tighter collaboration. Bombardieri and Bowman are the core authors and act as bridges, connecting multiple subfields or research teams.

Although Bowman from the UK leads in publication output with 21 papers, Se-

Table I. The top 10 productive and co-cited authors with publications concerning QoL in SjD.

Rank	Author	Affiliations and countries	Number of publications	Co-cited author	Affiliations and countries	Citations
1	Bowman Simon J	University Hospitals Birmingham NHS Foundation Trust, Rheumatology Department, England	21	Seror Raphaele	University of Paris-Saclay, Center for Immunology of Viral Infections and Autoimmune Diseases, Assistance Publique - Hôpitaux de Paris (AP-HP), Department of Rheumatology, France	326
2	Bootsma Hendrika	University of Groningen, Department of Rheumatology and Clinical Immunology, Netherlands	19	Vitali Claudio	Istituto Santo Stefano, Italy	263
3	Ng Wan-Fai	Newcastle University, Institute of Cellular Medicine, Musculoskeletal Research Group, England	15	Ramos Casals Manuel	Institute Clinic of Medicine and Dermatology (ICMiD), Department of Autoimmune Diseases, Spain	188
3	Vissink Arjan	University of Groningen, Department of Oral and Maxillofacial Surgery, Netherlands	11	Bowman Simon J	University Hospitals Birmingham NHS Foundation Trust, Rheumatology Department, UK	186
3	Jensen Janicke Liaaen	University of Oslo, Department of Oral Surgery and Oral Medicine, Norway	9	Fox Robert L	Scripps Memorial Hospital and Research Foundation, Rheumatology Clinic, USA	118
6	Baldini Chiara	University of Pisa, Rheumatology Unit, Italy	8	Meijer Jiska M	General Practitioners Research Institute, Netherlands	117
6	Mariette Xavier	Hôpital Bicêtre, Assistance Publique - Hôpitaux de Paris (AP-HP), Department of Rheumatology, France	8	Segal Barbara	University of Minnesota, USA	110
6	Palm Oyvind	Oslo University Hospital, Department of Rheumatology, Norway	8	Mariette Xavier	University of Paris-Saclay, Center for Immunology of Viral Infections and Autoimmune Diseases, Assistance Publique - Hôpitaux de Paris (AP-HP), Department of Rheumatology, France	101
6	Seror Raphaele	Hôpital Bicêtre, Assistance Publique - Hôpitaux de Paris (AP-HP), Department of Rheumatology, France	8	Shiboski Caroline H	Department of Orofacial Sciences, School of Dentistry, University of California, USA	95
10	Arends Suzanne	University of Groningen, Department of Rheumatology and Clinical Immunology, Netherlands	7	Brito Zeron Pilar	Institute of Biomedical Research August Pi i Sunyer (IDIBAPS), Spain	91

ror from the University of Paris-Saclay, Centre for Immunology of Viral Infections and Autoimmune Diseases, Hôpital Bicêtre, Assistance Publique, Hôpitaux de Paris (AP-HP), Department of Rheumatology, France, leads in citations with 326, followed by Vitali from Istituto Santo Stefano, Villa San Giuseppe, Italy, with 263 citations, and Ramos Casals from Institute Clinic of Medicine and Dermatology (ICMiD), Department of Autoimmune Diseases, Barcelona, Spain, with 188 citations. Among the top ten most-cited authors, there are 3 from the USA, two from France and Spain, indicating these countries' significant influence in the field (Table I).

Journal analysis

The 336 related articles were published in 151 journals. A visual analysis was conducted to showcase influential journals in the field. Among the publishing journals, the journal of *Clinical and Experimental Rheumatology* leads with the highest number of related publications (n=36, 10.71%), followed by *Rheumatology* (Oxford) (n=23, 6.85%), and the journal of *Rheumatology International* (n=16, 4.76%). The top ten journals collectively published 126 related articles, accounting for 37.5% of the total number of articles, indicating a significant concentration effect. In terms of citations, the journal of *Annals of the Rheumatic Diseases* leads with 1585 citations, followed by the journal of *Arthritis & Rheumatology* with 1122 citations, and *Rheumatology* (Oxford) with 990 citations. Looking at the top ten most-cited journals, five are ranked in the Q1 quartile and four in the Q2 quartile according to the *Journal Citation Reports* (JCR), demonstrating their significant impact in the field (Table II, Fig. 6).These journals, as leading specialty journals in the field of SjD, have played a pivotal role in disseminating and promoting research. Journal analysis can also assist researchers in rapidly identifying appropriate target journals for submission, thereby saving time in the journal selection process.

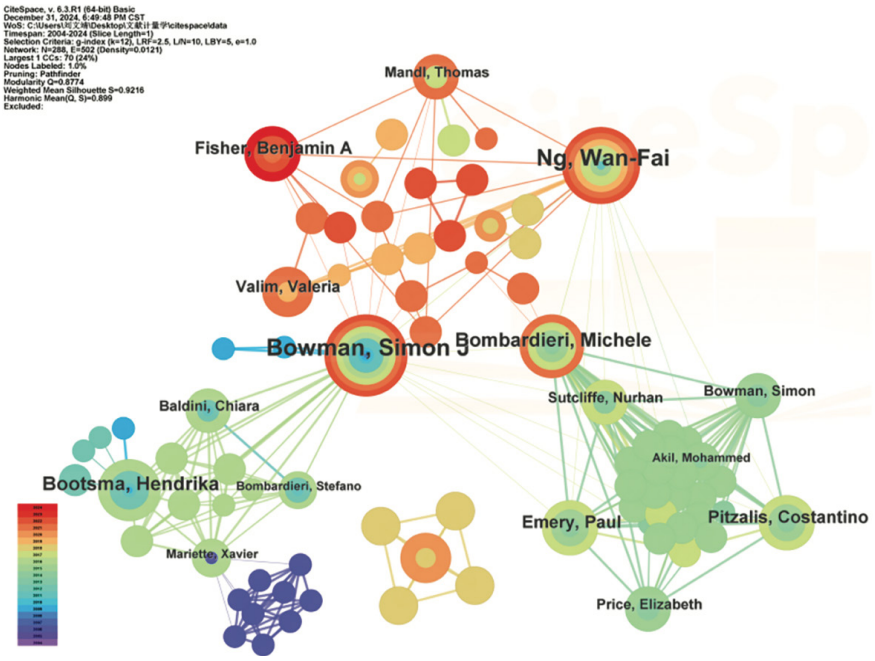

Fig. 5. The co-occurrence authors' map of QoL in SjD.

Reference analysis

Table III lists the top ten most-cited references related to quality of life in SjD. The most cited is the 2016 American College of Rheumatology/European League Against Rheumatism classification criteria for SjD, developed a single set of data-driven consensus classification criteria for primary SjD, which performed well in validation analyses and are well suited as criteria for enrolment in clinical trials (62). Secondly, the EULAR Sjögren's Syndrome Disease Activity Index (ESSDAI) for patients with primary Sjögren's syndrome, developed by the European League Against Rheumatism (EULAR) in 2010 (63), and Epidemiology of SjD by Qin *et al*. (64). Five out of the top ten most-cited articles were published in the *Annals of the Rheumatic Diseases*, with two each in *Rheumatology* (Oxford). This further underscores the central position of these journals within the field. Among these references, two studies specifically examined the health-related quality of life, employment status, and disability in SjD patients, revealing the significant impact of the disease on patients' daily life and social function (rank 5). Other studies explore predictors of health quality such as fatigue and mental health issues (rank 6), making them important references for both research and clinical practice.

Figure 7A displays the top ten cited references in the Web of Science core collection, with different colours representing the publication time of the articles, the closer to red, the more recent the publication, and the closer to purple, the older. Figure 7B shows the 25 articles with the highest citation burst intensity. The earliest burst started in 2004, with the highest burst intensity being 11.5 for Shiboski *et al*. 2017 article in *Annals of the Rheumatic Diseases* on "2016 American College of Rheumatology/European League Against Rheumatism classification criteria for primary Sjögren's syndrome" (62). The years 2008, 2010, and 2016-2022 witnessed more frequent high-citation bursts, indicating a heightened focus on quality-of-life research in SjD during these years.

Table II. The top 10 most productive journals and co-cited journals concerning QoL in SjD.

Rank	Journals	Number of publications	Number of citations	Average number of citations	IF (2023), JCR quantile	Co-cited journals	Number of co- citations	Centrality	IF (2023), JCR quantile
1	Clinical and Experimental Rheumatology	36	639	17.75	3.5, Q2	Annals of the Rheumatic Diseases	1585	0.01	20.3, Q1
2	Rheumatology (Oxford)	23	1306	56.78	4.7, Q1	Arthritis & Rheumatology	1122	0.01	11.4, Q1
3	Rheumatology International	16	395	24.69	3.2, Q2	Rheumatology (Oxford)	990	0.01	4.7, Q1
4	Journal of Rheumatology	10	240	24	3.6, Q2	Journal of Rheumatology	651	0.02	3.6, Q2
5	Journal of Clinical Medicine	10	135	13.5	3.0, Q1	Clinical and Experimental Rheumatology	642	0.06	3.5, Q2
6	Clinical Rheumatology	8	84	10.5	2.9, Q2	Arthritis Care & Research	360	0.04	3.7, Q1
7	PLoS One	7	250	35.71	2.9, Q1	Scandinavian Journal of Rheumatology	296	0.02	2.2, Q3
8	Arthritis Care & Research	6	221	36.83	3.7, Q1	PLoS One	206	0.02	2.9, Q1
9	RMD Open	5	44	8.8	5.1, Q1	Rheumatology International	195	0.04	3.2, Q2
10	Annals of the Rheumatic Diseases	5	522	104.4	20.3, Q1	Clinical Rheumatology	180	0.07	2.9, Q2

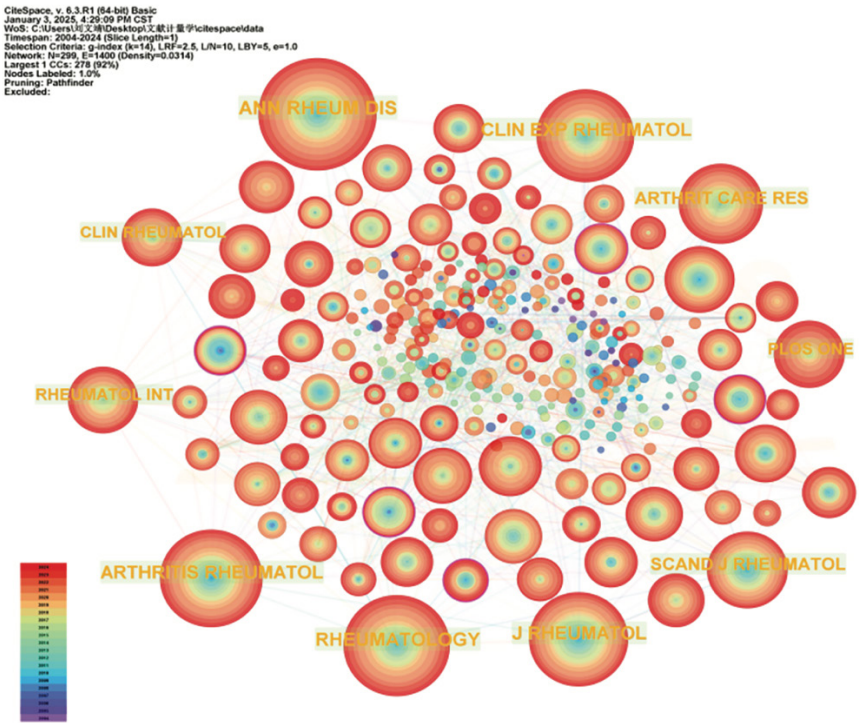

Fig. 6. The co-cited journals' map of QoL in SjD.

Keywords and hotspot analysis

Keyword analysis quickly identifies the research content and hotspots within the field. As shown in Figure 8, except for Sjogren syndrome, the most common keyword is "quality of life", appearing 80 times, which reflects the central focus of the research. The five keywords are "fatigue", "xerostomia", "depression", "sexual dysfunction" and "xerophthalmia", as well as "health-related quality of life", indicating that the research focuses more on the significant impact of common symptoms on the quality of life of SjD patients. The frequent occurrence of "health-related quality of life" suggests that the research places greater emphasis on patients' self-perception and self-reports. This highlights the need for a comprehensive approach to the management of SjD, not only focusing on treating the disease but also on improving patients' quality of life and mental health. In the analysis of keyword burst intensity, terms like "classification", "primary sjogrens syndrome", "consensus", "systemic lupus erythematosus" and "rheumatoid arthritis" are among the top in burst strength. Keyword and hotspot analysis can reveal the frontier topics in this research field. Overall, global studies on the QoL of patients with SjD have been steadily increasing, shifting from an early focus on fundamental theories and disease classification towards research centred on patient experiences. Such studies

Table III. The top 10 cited references concerning QoL in SjD.

Rank	Title	First author	Year	Journal	Citations in Google scholar
1	2016 American College of Rheumatology/European League Against Rheumatism classification criteria for primary Sjögren's syndrome	Shiboski *et al.*	2016	Annals of the Rheumatic Diseases	2715
2	EULAR Sjögren's syndrome disease activity index: development of a consensus systemic disease activity index for primary Sjögren's syndrome	Seror *et al.*	2010	Annals of the Rheumatic Diseases	1031
3	Epidemiology of primary Sjögren's syndrome: a systematic review and meta-analysis	Qin *et al.*	2015	Annals of the Rheumatic Diseases	903
4	EULAR Sjögren's Syndrome Patient Reported Index (ESSPRI): development of a consensus patient index for primary Sjögren's syndrome	Seror *et al.*	2011	Annals of the Rheumatic Diseases	554
5	Health-related quality of life, employment and disability in patients with Sjögren's syndrome	Meijer *et al.*	2009	Rheumatology (Oxford)	316
6	Primary Sjögren's syndrome: health experiences and predictors of health quality among patients in the United States	Segal *et al.*	2009	Health and Quality of Life Outcomes	271
7	Measurement of fatigue and discomfort in primary Sjogren's syndrome using a new questionnaire tool	Bowman *et al.*	2004	Rheumatology (Oxford)	258
8	Health-related quality of life in primary Sjögren's syndrome rheumatoid arthritis and fibromyalgia compared to normal population data using SF-36	Strömbeck *et al.*	2000	Scandinavian Journal of Rheumatology	188
9	Sjögren's syndrome	Fox *et al.*	2005	The Lancet	130
10	Classification criteria for Sjögren's syndrome: a revised version of the European criteria proposed by the American-European Consensus Group	Vitali *et al.*	2002	Annals of the Rheumatic Diseases	126

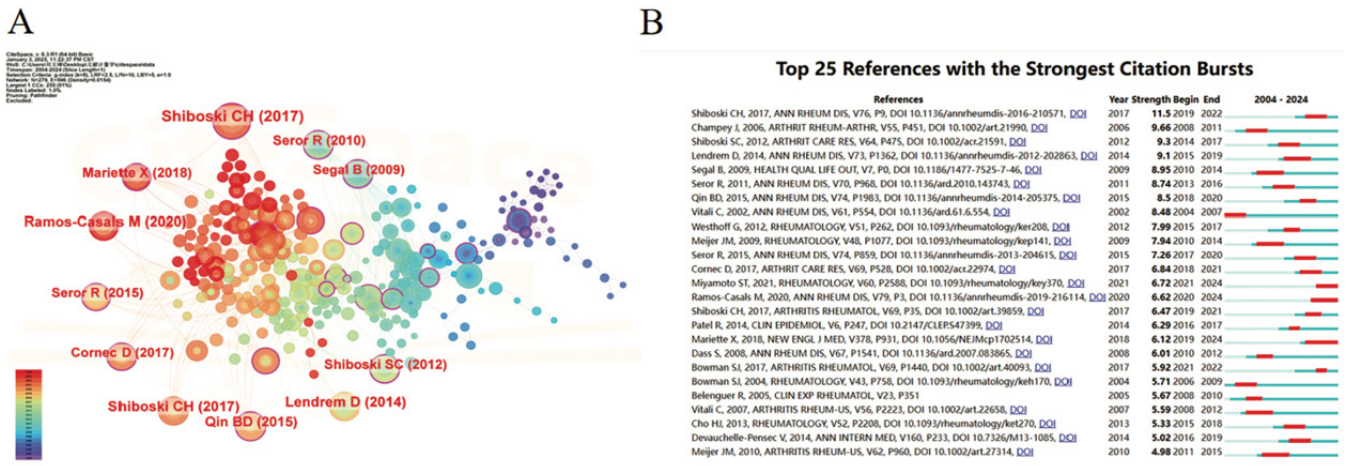

Fig. 7. **A**: The analysis of references related to QoL in SjD. **B**: The top 25 references with strong citation bursts.

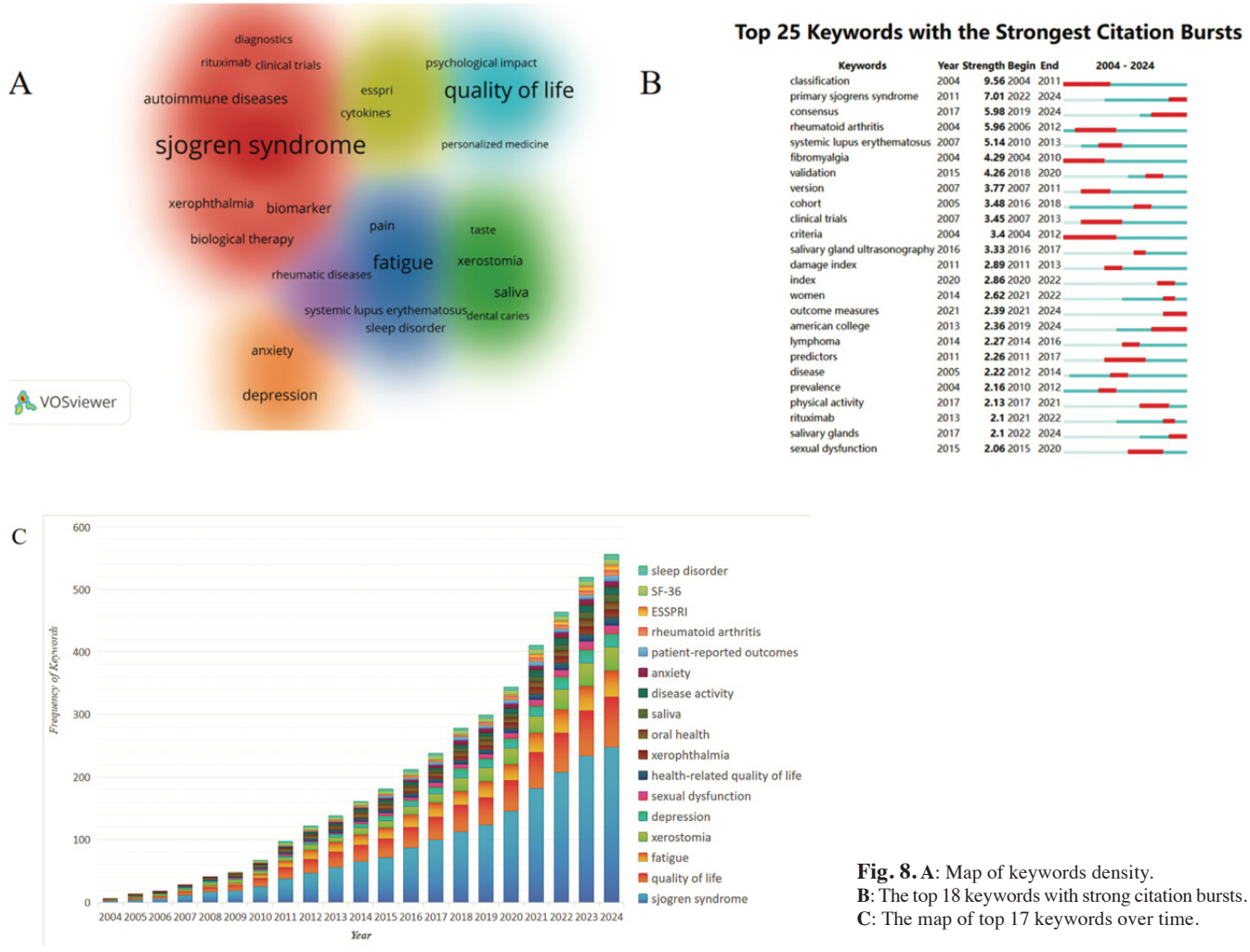

Fig. 8. **A**: Map of keywords density.
B: The top 18 keywords with strong citation bursts.
C: The map of top 17 keywords over time.

include investigations into the impact of dry eyes and mouth, fatigue, anxiety and depression, and sexual dysfunction on QoL; the application of tools such as ESSPRI and SF-36 in QoL assessment; and research on biomarkers and personalised treatments. The scope of this work spans from the development of assessment tools, analysis of influencing factors, and evaluation of mental health to the exploration of therapeutic approaches. This trend marks a profound shift in research emphasis from 'the disease itself' to 'the patient's experience', with the goal of achieving more precise management and interventions.

Discussion

Overview of research progress in the field

This study is the first to comprehen-

sively analyse research on the QoL of patients with SjD using bibliometric methods based on the Web of Science Core Collection (WoSCC) from 2004 to 2024. According to the data retrieved from WoSCC, between 2004 and 2024, a total of 922 authors from 56 countries and 630 institutions published 336 articles on SjD and QoL in 151 journals. The annual number of publications has shown an overall upward trend, indicating that this research field has received increasing attention from scholars in recent years. We predict that the field holds substantial growth potential, with a growing number of countries beginning to focus on QoL in SjD patients. However, as a disabling autoimmune disease that severely impacts patients' lives, SjD has received disproportionately limited dedicated QoL research in terms of publication volume and growth rate relative to its clinical significance. Recent clinical reviews have further emphasised that SjD is a complex systemic autoimmune condition, with Sicca symptoms, systemic manifestations, and comorbidities profoundly impairing patients' quality of life, and highlighted the urgent need for innovative treatment strategies (65). Several factors may account for this: the subjective and heterogeneous nature of SjD symptoms poses challenges for standardised research; funding and attention in this field may be more heavily directed toward exploring disease mechanisms and developing classification criteria rather than patient-reported outcomes (PROs); and compared to other autoimmune diseases such as rheumatoid arthritis, the lack of effective targeted therapies for SjD may indirectly reduce enthusiasm for investigating how treatment can improve QoL.

In-depth analysis of collaboration networks among countries, institutions, and authors revealed a clear trend in which the field is concentrated around a small number of core teams, gradually expanding toward international collaboration. The United Kingdom ranks highest in both publication volume and citations in SjD QoL research, engaging in extensive collaboration with multiple countries worldwide. Notably, among the top five countries in publication output, four are developed countries and one China is a developing country. Among the top ten countries, developed and developing countries are equally represented (five each). Although we could not assess the correlation between per capita GDP and publication volume, total GDP serves as an indicator of a nation's overall economic output and directly influences its capacity to fund research. It is evident that countries with higher GDP are more capable of focusing on QoL, whereas developing countries and regions still need to strengthen cooperation and exchanges with these developed nations to improve QoL for SjD patients.

Research hotspots addressing clinical challenges: keywords reveal the significance of unmet clinical needs

High-frequency keywords such as "fatigue", "depression", and "sexual dysfunction" reflect the core challenges and unmet clinical needs in the current management of SjD. These symptoms are directly associated with patients' subjective experiences and daily functioning, yet are often marginalised in clinical trials and guidelines, lacking systematic intervention strategies.

Among them, "fatigue" emerged as the most prominent keyword, underpinned by complex and multifactorial mechanisms involving inflammatory, neuroendocrine, and psychosocial pathways. Recent studies have reported that SjD-related fatigue is closely associated with abnormalities in the tryptophan kynurenine metabolic pathway, with the kynurenine/tryptophan ratio showing the strongest correlation with fatigue severity, suggesting that immune inflammation driven metabolic alterations may represent an important underlying pathophysiological mechanism (66). Other studies have identified associations between fatigue and biomedical indicators such as reduced leukocyte counts or elevated immunoglobulin G levels; however, fatigue often persists even after correction of these abnormalities (67). Fatigue is prevalent across different clinical subtypes of SjD, yet current treatment efficacy remains limited, frequently failing to meet the expectations of both patients and clinicians highlighting the urgent, unmet clinical need stemming from this high-prevalence yet low-treatment-response paradox (68).

The high frequency of "depression" and "anxiety" indicates that SjD is not merely a somatic disease but a prototypical psychosomatic disorder. Chronic pain, functional impairment, and social isolation collectively contribute to a substantial psychological burden on patients. This further supports the notion that SjD-related fatigue likely has a dual nature arising from both biomedical mechanisms (particularly immune dysregulation) and psychosocial influences (69). Nevertheless, routine clinical practice often lacks systematic screening and integrated interventions for mental health. "Sexual dysfunction", as a long-overlooked yet QoL-impairing dimension, highlights an area of taboo and neglect in both clinical inquiry and research. The clustering of these keywords not only reflects the genuine suffering of patients but also reveals the limitations of the current biomedical model in addressing a complex condition such as SjD. Therefore, there is an urgent need for a systematic review and integrated evaluation of the biomedical and psychosocial factors contributing to SjD-related symptoms, in order to explore their independent effects, interactions, and potential self-reinforcing mechanisms. Such a research approach will not only help clarify existing gaps in treatment but also provide a scientific basis for developing more individualised and outcome-optimised intervention strategies strategies that are likely to require the integrative application of multimodal therapeutic approaches.

Paradigm shift: evolution from disease definition to patient-centred care

The evolution of keyword timelines clearly reflects the continuous deepening of research in the field of SjD and QoL. Over time, the research focus has exhibited distinct stage-specific characteristics: the early stage (2004-2011) centred on fundamental definitions and disease classification; the intermediate stage (2012-2018) focused on tech-

nological advancements and clinical validation; and the most recent stage (2019-2024) has shifted towards treatment optimisation and enhancement of QoL. This progression illustrates the transition of the field from theoretical framework building to clinical practice, and further towards addressing the comprehensive needs of patients. The focus of research has gradually expanded from defining the disease itself to optimising treatment strategies and improving QoL, reflecting an emerging tendency in both academia and clinical practice to strike a balance between elucidating disease mechanisms and improving patient outcomes.

Our reference analysis reveals a noteworthy 'dual narrative'. On the one hand, the most highly cited papers are primarily concentrated on disease classification criteria (62), disease activity assessment tools (63), and epidemiological studies (64). These 'cornerstone publications' have been extensively cited in the majority of SjD-related studies, forming the foundational knowledge base of the field. On the other hand, burst citation analysis enabled us to identify 'core publications' that have played pivotal roles in advancing the QoL subfield of SjD research papers whose total citation counts may not be the highest overall but have exerted a decisive influence on thematic development. For example, Miyamoto *et al*. (70) provided a comprehensive review summarising the impact of multiple symptom domains, health-related quality of life assessment methods, and the socioeconomic burden of SjD; and Lendrem *et al*. (71) demonstrated that pain and fatigue are major determinants of disease burden and QoL in primary SjD. Collectively, these publications reflect a critical shift in the field towards greater recognition of patient-reported outcomes and the development of symptom-specific interventions. Qualitative analysis of these core references indicates that QoL research has undergone three major evolutionary shifts: 1. moving from global QoL evaluation to in-depth exploration of specific dimensions such as sexual function and mental health; 2. progressing from identifying associated factors to investigating feasible interventions; and 3. advancing from the application of general assessment tools to the design and adoption of SjD-specific instruments. The analytical strategy employed in this study highlights important contributions that, despite not having high overall citation frequencies, have played a leading role in shaping the direction of QoL research. These findings provide subsequent researchers with a more precise and targeted knowledge foundation for advancing the field.

Future directions

Based on the above analysis, we propose the following directions for future research: 1. Mechanistic research: conduct in-depth studies into the biological mechanisms underlying core symptoms such as fatigue and pain, with the aim of identifying effective therapeutic targets; 2. Clinical practice: establish multidisciplinary management models integrating rheumatology, stomatology, ophthalmology, and psychiatry/psychology, and incorporate routine mental health screening into clinical care; 3. Methodology: Encourage the use of digital health technologies (*e.g*. mobile applications) to collect patient-reported outcomes (PROs) in real time, thereby overcoming the limitations of retrospective questionnaires; 4. International collaboration: Strengthen multicentre collaborations between developed and developing countries, and conduct large-scale, long-term cohort studies to ensure that research on and clinical management of QoL in SjD patients is globally representative.

Limitations

In this study, we selected the Web of Science Core Collection (WoSCC) as the sole data source, primarily due to its high level of data standardisation, well-structured citation information, and excellent compatibility with bibliometric analysis tools. The WoSCC has high authority in indexing high-quality international journals and covers the vast majority of core English-language journals also indexed in Scopus and PubMed. Although Scopus and PubMed offer certain complementary coverage for example, by including more conference papers and regional journals their citation data structures are generally less complete than those of WoSCC, and some of their data formats are less compatible with specific bibliometric tools. Therefore, WoSCC was prioritised in this study to ensure the accuracy and comparability of the analysis. Nevertheless, we acknowledge that future studies could integrate WoSCC, Scopus, and PubMed data to obtain more comprehensive literature coverage.

Conclusion

This study, through bibliometric analysis, comprehensively elucidates the developmental trends, knowledge structure, and evolutionary patterns of research on the QoL of patients with SjD. Our findings indicate that the field is currently at a critical transitional stage, shifting from a focus on 'disease cognition' to the deeper paradigm of 'patient-centred care', characterised by a stable yet slow growth trajectory. High-frequency keywords such as "fatigue", "depression" and "sexual dysfunction" are not merely descriptors of symptoms but constitute a strong call for future research calling for more effective therapeutic approaches, more comprehensive evaluation perspectives, and more humane models of care. Systematically addressing and improving the QoL of SjD patients will be a crucial step in advancing rheumatology research from 'curing the disease' to 'healing the patient'.

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