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Original Article

A scientometric and comparative study of Sjogren's syndrome research by rheumatologists and stomatologists

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Abstract *Background/purpose:* The diagnosis and treatment of Sjogren's syndrome (SS) are commonly conducted by rheumatologists and stomatologists. The purpose of this study was to compare the scientometric characteristics of SS publications by rheumatologists and stomatologists.
Materials and methods: All the papers on cheilitis were comprehensively retrieved from the Scopus database, and divided into rheumatologists and stomatologists groups.
Results: There were 3245 and 1209 papers on SS were published by rheumatologists and stomatologists, respectively. For the most-cited top-200 papers, the total citation count was 29,764 and the *h* index was 108 for SS publications by rheumatologists; whereas the count is 19,891 and *h* index is 81 for publications by stomatologists. Interestingly, we observed that accumulated citations of the publications by stomatologists cooperated with rheumatologists were larger than those by stomatologists alone during 2012–2022. The more common keywords such as saliva, salivation, minor salivary glands, parotid gland, submandibular gland, sialography, lip, dental caries, and hyposalivation were reported by stomatologists. The more frequent

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keywords such as rheumatoid factor, fatigue, lymphoma, interstitial lung disease, arthralgia, Raynaud phenomenon, lymphadenopathy, and vasculitis were reported by rheumatologists.
Conclusion: This study firstly reports the scientometric characteristics of SS publications by rheumatologists and stomatologists. The scale and citations of rheumatologists' publications greatly outweigh those of stomatologists, suggesting stomatologists can cooperate more with rheumatologists regarding SS research.

Introduction

Sjogren's syndrome (SS) as the second most common autoimmune disease ranking next to rheumatoid arthritis is characterized by dysfunction and destruction of exocrine or secretory glands, particularly the salivary and lacrimal glands.[1] SS can exist as either a primary syndrome or as a secondary syndrome associated with other autoimmune diseases, such as rheumatoid arthritis, systemic lupus erythematosus, systemic sclerosis and scleroderma.[1] Both primary and secondary SS share the similar cardinal signs and symptoms, xerostomia and xerophthalmia; generally, both are treated similarly.[2] Early diagnosis and treatment are important to prevent the complications of SS; but the diagnosis is often delayed for 5 or even more years owing to the insidious onset of this disease.[2] The therapeutic management of SS has not improved substantially in recent decades, and therapeutic strategies remain challenging in clinical practice. Currently no definitive test or cure exists, and treatment is predominately palliative and supportive.[2,3]

Generally, the diagnosis and treatment of SS are conducted by stomatologists and rheumatologists. SS patients often present first to their oral health professional because of their predominantly oral symptomatology, mainly xerostomia.[4,5] Hence, stomatologists are frequently the first practitioners to detect the signs of SS, and then rheumatologists are usually the practitioners to manage the systemic symptomatology, such as fatigue, lymphoma, interstitial lung disease, arthralgia, and Raynaud phenomenon, in the middle and late stages of the disease.[6–8] Given the complex and numerous challenges of diagnosing and treating this syndrome, increasingly large number of papers regarding SS have been published. The papers published by rheumatologists and stomatologists preferentially represent the scientific output and concerns of the same disease. Scientometrics is a useful tool that utilizes bibliometric and citation data to assess scientific output within the designated area.[9–12]

Herein, we hypothesized that there might be different scientific output of SS research by rheumatologists and stomatologists. Therefore, the purpose of this study was to compare the scientometric characteristics of SS publications by rheumatologists and stomatologists, so as to promote mutual understanding and even reciprocal cooperation regarding this syndrome in rheumatology and stomatology.

Materials and methods

Based on the methodology described previously,[9–11] we searched the literature up to 30 Aug 2023 from the Scopus database according to the search strategy (Table S1). We used medical subject term "Sjogren*" OR "Sjögren*" in the Title to retrieve all the papers on SS, without restriction to language, type, and year of publication. In literature search, the asterisk indicates a wildcard used to search for all endings including fifth or more root words. In clinical practice, rheumatologists and stomatologists generally belong to the rheumatology and stomatology affiliation, respectively. Hence, the papers with the word ("rheumat*") and (dent* OR oral OR stomatolog*) in the affiliation generally represent scientific output of rheumatologists and stomatologists, respectively. The papers with both 2 words concurrently in the affiliation represent scientific output of their cooperation. Then, SS publications by rheumatologists, stomatologists, and their cooperation were retrieved, respectively. The scientometric characteristics of all the eligible articles were reviewed and recorded the following information: publication year, title, keywords, citation count, paper type, authorship, affiliation, and country/region of origin. Data search and extraction were performed independently by two investigators (H.S. and W.L.), and discrepancy of results was resolved in a consensus symposium. The Bibliometrix Biblioshiny R-package software (https://www.bibliometrix.org/home/; K-Synth Srl Inc., Naples, Italy) was used to analyze the relevant bibliometric data.

Results

Citation characteristics

With the search strategy algorithm, a total of 3245 and 1209 papers on SS were published by rheumatologists and stomatologists (with a ratio, 2.7:1), respectively. There were 591 publications by rheumatologists cooperated with stomatologists. Fig. 1A illustrates the number and distribution of the paper types. To assess scientific influence of the academics, the most-cited top-200 papers on SS were retrieved. The total citation count was 29,764 and the *h* index was 108 for SS publications by rheumatologists, and the total count was 19,891 and the *h* index was 81 for SS publications by stomatologists (Fig. 1B).

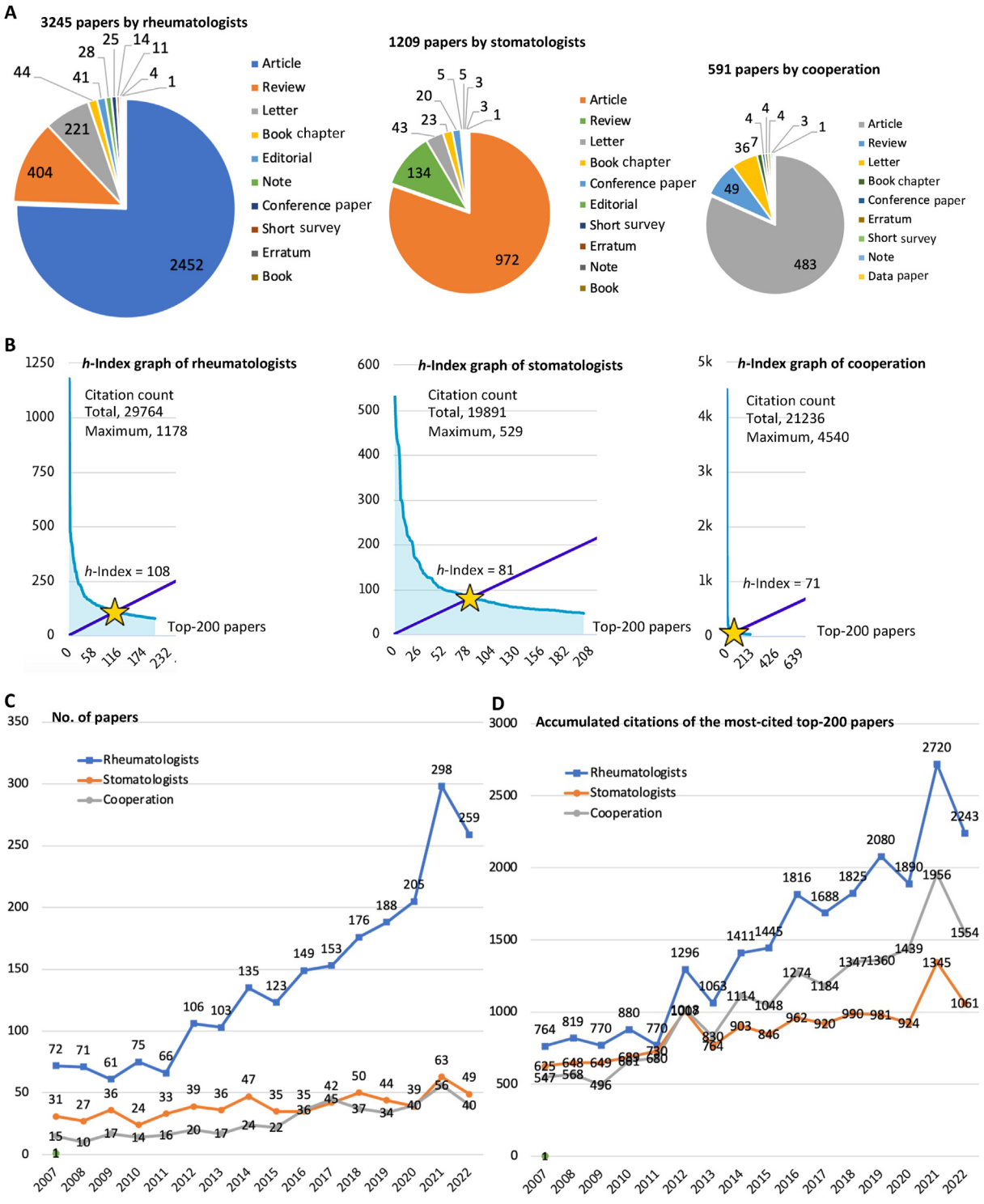

Figure 1 Citation characteristics of the papers on Sjogren's syndrome. (**A**) Document types and distribution of the papers. (**B**) The *h*-Index graphs of the most-cited top-200 papers. (**C**) The annual number of the papers during 2007–2022. (**D**) The accumulated citations of the most-cited top-200 papers during 2007–2022.

Besides, the total count was 21,236 and the *h* index was 71 for SS publications by rheumatologists cooperated with stomatologists. The detailed information on publication year, title, journal, citation count, authors, affiliation, keywords, and document types of the top-200 papers are presented in Table S2-S4.

To further concretize the treads of scientific output in SS research field, we assessed the annual number of the papers and accumulated citation count of the most-cited top-200 papers during 2007 to 2022. The annual number of the publications by rheumatologists stably raised from 72 to 298 during 2007–2021 (Fig. 1C), and the accumulated citations

of the top-200 papers stably increased from 764 to 2720 during this period (Fig. 1D). The annual number of the publications by stomatologists raised from 31 to 63 during 2007—2021, and the accumulated citations of the top-200 papers increased from 625 to 1956 during this period. Interestingly, the number of publications by stomatologists cooperated with rheumatologists was modest growth from 15 to 56 during 2007—2021; while the accumulated citations of the top-200 papers stably increased from 547 to 1345. However, all these numbers were decreased in varying degrees in 2022 compared to 2021.

Bibliometric characteristics

The cloud graphs of journal names, contributing authors, institutions, and countries/regions are showed in Fig. 2. For rheumatologists, the journal with largest number was *Clinical & Experimental Rheumatology* (n = 235), followed by *Annals of The Rheumatic Diseases* (n = 142) and *Clinical Rheumatology* (n = 130). The contributing author with largest number of papers was Baldini, C. (n = 88), followed by De Vita, S. (n = 87) and Mariette, X. (n = 85). The contributing institution with the maximum number was Università di Pisa (n = 103), followed by Universität zu Lübeck (n = 100) and Peking Union Medical College Hospital (n = 96). The country of origin with largest number of papers was China (n = 506), followed by United States (n = 480) and Italy (n = 408).

For stomatologists, the journal with largest number was *Oral Surgery Oral Medicine Oral Pathology Oral Radiology* (n = 55), followed by *Oral Diseases* (n = 47) and *Journal of Oral Pathology & Medicine* (n = 33). The contributing author with largest number of papers was Hayashi, Y. (n = 56), followed by Saito, I. (n = 40) and Tsubota, K. (n = 39). The contributing institution with the maximum number was National Institute of Dental and Craniofacial Research (n = 85), followed by National Institutes of Health (n = 82) and Tokushima University (n = 74). The country of origin with largest number of papers was United States (n = 422), followed by China (n = 318) and Japan (n = 110).

Figure 2 Cloud graphs of (**A**) journal of publication, (**B**) contributing authors, (**C**) institutions, and (**D**) countries/regions of origin in the publications by rheumatologists, stomatologists, and their cooperation, respectively.

Research characteristics

Based on the frequency of keywords in all included papers, we highlighted the analysis of research characteristics of SS publications by rheumatologists and stomatologists (Fig. 3A). All the keywords were automatically recognized in the order of highest to lowest frequency by the database. The related disorders, research keywords, study design, and drug research were identified. The research keywords such as salivary gland, immunology, pathology, pathogenesis,

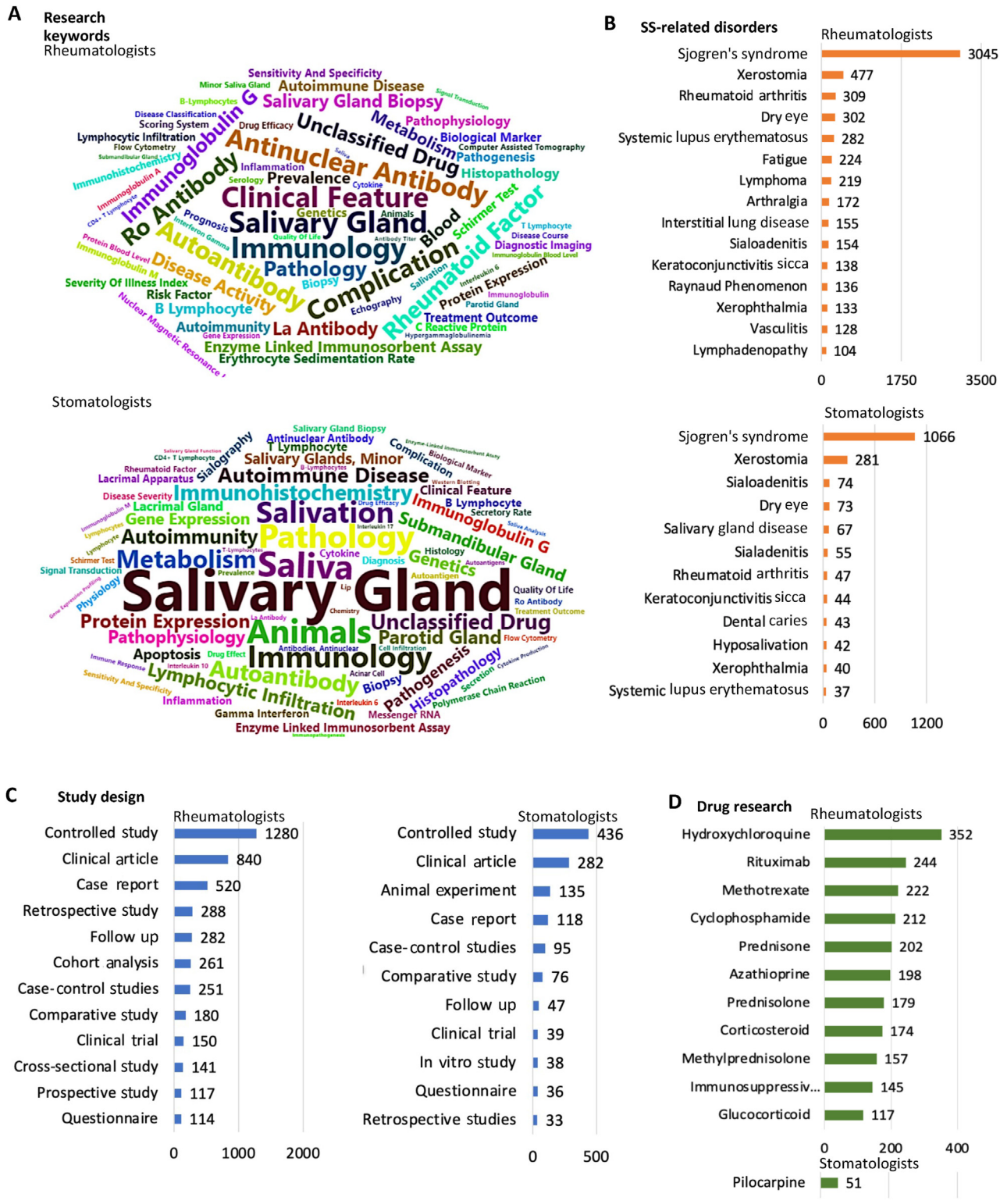

Figure 3 Research characteristics of the papers on Sjogren's syndrome (SS). (**A**) Cloud graphs of research keywords. The ranks of (**B**) SS-related disorders, (**C**) study design, (**D**) drug research.

autoantibody, antinuclear antibody, autoimmunity, B lymphocyte, immunoglobulin G, metabolism, and protein expression were relatively common in the publications by both rheumatologists and stomatologists. For rheumatologists, the keywords such as complication, rheumatoid factor, disease activity, prevalence, erythrocyte sedimentation rate, risk factor, and schirmer test were more common in their publications in comparison with those publications by stomatologists. For stomatologists, the keywords such as saliva, salivation, minor salivary glands, parotid gland, submandibular gland, lacrimal gland, sialography, apoptosis, lip, and animals were more common in their publications in comparison to those publications by rheumatologists.

The most of the SS-related disorders such as xerostomia, sialoadenitis, rheumatoid arthritis, keratoconjunctivitis sicca, and dry eye were similar in the publications by both rheumatologists and stomatologists. The distinctive disorders such as fatigue, lymphoma, interstitial lung disease, arthralgia, Raynaud phenomenon, lymphadenopathy, and vasculitis were mainly concerned by rheumatologists; whereas the distinctive disorders such as salivary gland disease, dental caries, and hyposalivation were mainly concerned by stomatologists (Fig. 3B). The most of the study design such as controlled study, clinical article, case report, case—control studies and follow up were similar in the publications by both rheumatologists and stomatologists. The cohort analysis, cross-sectional study, and prospective study were mainly performed by rheumatologists; whereas animal experiment and in vitro study were mainly conducted by stomatologists (Fig. 3C). Interestingly, the drugs such as hydroxychloroquine, rituximab, methotrexate, cyclophosphamide, prednisone, azathioprine, and corticosteroid were commonly reported in the publications by rheumatologists; but only pilocarpine was commonly reported in the publications by stomatologists (Fig. 3D).

Discussion

Although immunological and histopathological evidence support for an autoimmune pathogenesis, the etiology and pathogenesis of SS have not been fully elucidated.[13,14] As a systemic autoimmune disease, SS is characterized by hyperreactivity of B lymphocytes and by the production of various antibodies, which causes dysfunction and destruction of multiple exocrine glands, especially salivary and lacrimal glands.[1] Diagnosis of SS is based on the concurrent presence of various signs and symptoms as established by 6 aspects set by the American European Consensus Group standards: oral symptoms, ocular symptoms, evidence of oral signs, evidence of ocular dryness, evidence of salivary gland involvement with positive Anti-Ro/La (SSA/SSB) autoantibodies, and a positive gland biopsy.[15] Currently, labial salivary gland biopsy is widely accepted as the standard method to diagnose SS due to a high disease specificity and limited invasiveness.[16] As expected, we observed that the common keywords such as salivary gland, immunology, autoimmunity, pathogenesis, autoantibody, B lymphocytes, antinuclear (Ro/La) antibody, and biopsy are reasonable for SS research.

The first and foremost symptom of SS is xerostomia, which represents a clinical condition characterized by salivary flow rate decrease.[6] Low salivary flow does not permit oral self-cleansing which buffers, lubricates and performs essential antimicrobial duties, results in multiple oral manifestations and complications such as dental caries, periodontal disease, oral candidiasis, dysphagia, dysgeusia, halitosis.[6,17] Besides, SS often interests major salivary gland involvement, mainly parotid gland swelling, but also isolated submandibular gland swelling. Accordingly, we observed that the more common keywords such as saliva, salivation, minor salivary glands, parotid gland, submandibular gland, sialography, lip, dental caries, and hyposalivation are reasonable for SS research by stomatologists. Subsequently, SS can cause a myriad of systemic symptoms and complications, owing to exocrine gland dysfunction, enhanced production of rheumatoid factors, and lymphocytic infiltration of multiple organs, including musculoskeletal, hematologic, gastrointestinal, pulmonary, and renal complications, further reduce the quality of life of patients.[18] Hence, we observed that the more frequent keywords such as rheumatoid factor, fatigue, lymphoma, interstitial lung disease, arthralgia, Raynaud phenomenon, lymphadenopathy, and vasculitis are reasonable for SS research by rheumatologists.

Since SS has no cure, treatment is symptomatic and supportive, starting with the management of the central triplet of symptoms (dryness, fatigue and pain) followed by the management of systemic disease.[13] We observed that pilocarpine was commonly reported by stomatologists. This drug alleviates the oral and ocular symptoms related to xerostomia and xerophthalmia by increasing exocrine glands secretion, but it can also give rise to adverse effects which reflects its other cholinergic actions.[6] According to the 2019 European League Against Rheumatism (EULAR) evidence and consensus-based recommendations for the management of SS patients,[13] systemic medications contain oral muscarinic agonists (pilocarpine, cevimeline), oral glucocorticoids, hydroxychloroquine, synthetic immunosuppressive agents (methotrexate, cyclophosphamide, leflunomide, azathioprine, and mycophenolate), and biological therapies (abatacept, rituximab, and belimumab). EULAR recommendations will be broadly applied in clinical practice and/or serve as a template for national societies to develop local recommendations. We observed that the drugs such as prednisone, corticosteroid, hydroxychloroquine, azathioprine, methotrexate, cyclophosphamide, and rituximab were commonly reported by rheumatologists but not commonly done by stomatologists. Moreover, we also observed that various cells and cytokines are involved in SS, including immunoglobulins, T lymphocyte, B lymphocyte, interleukins, and interferon gamma (Fig. 3). Hence, targeting these cells and cytokines may help to modulate the immune response and improve SS treatment.

As mentioned above, stomatologists and rheumatologists are frequently the practitioners to manage oral and systemic symptomatology, respectively. Although the management of first signs by stomatologists are important to prevent the progression of SS, the management of middle and late stages with a long course of disease by rheumatologists are more complex and of more numerous challenges. It is important to establish a close relationship between the stomatologists and the rheumatologists in order to make an early and correct diagnosis, promoting appropriate hygiene and dietary measures, as well as to treat and prevent potential oral and systemic

complications. Understandably, we observed that the number and citations of the publications by rheumatologists were larger than those by stomatologists. Generally, the academic impact (impact factor) of rheumatology journals is higher than stomatology journals. Interestingly, we observed that accumulated citations of the publications by stomatologists cooperated with rheumatologists were also larger than those by stomatologists alone during 2012—2022, notwithstanding the annual number of the cooperated publications was lower than that by stomatologists alone (Fig. 1). Interprofessional collaboration and multidisciplinary management of underlying systemic conditions, such as SS, is imperative to help reduce complications and comorbidities.[7] When dental and medical professionals work together as a team, they can improve the outcomes and quality of life for the patient diagnosed with SS.

In summary, the scientometric characteristics of SS publications by rheumatologists and stomatologists are firstly comprehensively reported in this study. The scale and citations of rheumatologists' publications greatly outweigh those of stomatologists, suggesting stomatologists can cooperate more with rheumatologists regarding SS research. Such collaboration of care by the dental and medical professionals will improve the quality of life of the SS patients and lead to better patient outcomes.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jds.2024.01.020.

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