

Global trends on *Blastocystis* spp. research: A scientometric study

Original
Article

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ABSTRACT

Background: For many years, the classification and pathogenicity of *Blastocystis* was a subject of debate. The study rational is built on two hypotheses; whether this uncertainty affects the research on *Blastocystis* spp., and whether regional differences have an impact on diseases prevalence.

Objective: The aim of this study is to guide researchers interested in blastocystosis by presenting a bibliometric review of the existing literature on blastocystosis.

Material and Methods: The VOSviewer visualization methodology was used. Institutions, nations, international collaborations, journals, articles, authors, keywords, co-authors, co-citations, and citation rates from the WoS database formed the study data. A total of 1192 documents were found in the WoS database, of which 906 original articles were analyzed.

Results: The articles were cited 24,065 times in total and the mean Hirsch (H) index was 74. The countries with the highest number of articles were as follows: USA (n = 93; 10.26%), Singapore (n = 79; 8.72%), China (n = 76; 8.39%), and Malaysia (n: 74; 8.17%). During the period from 2000-2013, there were a limited number of articles, while since 2014 there was a minimum of 34 publications per year. The highest number of articles was published in 2021 (n = 87) and the highest number of citations (n=3,485) was in 2021. The leading affiliation according to the number of published articles was the National University of Singapore (8.72%), University Malaya (6.62%), and Nara Women's University (4.64%). The USA, Singapore, China and Malaysia have led scientific production on *Blastocystis* spp.

Conclusion: Due to development of recent molecular technology, the interest in *Blastocystis* spp. increased as recorded by ample publications and cited references. Development of interdisciplinary scientific research networks to include the most productive countries is crucial for *Blastocystis* related studies.

Keywords: *B. hominis*, bibliometrics, citation, irritable bowel syndrome, pathogenicity, zoonosis.

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INTRODUCTION

Blastocystis spp. is a common single-celled intestinal parasite belonging to the Stramenopile group^[1]. It has been determined that humans and several animal species carry this parasite as natural hosts, and certain subtypes of animals infect humans^[2]. Based on analyses of the small subunit rRNA gene (SSU rDNA) of *Blastocystis* spp, 22 subtypes have been identified in a variety of animals and in humans^[3]. The prevalence of the subtypes varies widely among regions^[4]. The prevalence of *Blastocystis* spp. in developing countries is much higher than in developed countries^[5]. It was reported that *Blastocystis* spp. is responsible for 30%–40% of diarrhea cases, especially in developing countries^[6]. The wide prevalence of blastocystosis and the increasing risk rates with age in these countries point toward waterborne transmission as well as human-

to-human transmission or zoonotic transmission. However, some studies reported that *Blastocystis* spp. rates in asymptomatic individuals are similar in both developing and developed countries^[7]. Other studies reported that *Blastocystis* spp. was associated with irritable bowel syndrome and urticaria^[8,9]. A higher prevalence of *Blastocystis* spp. was reported in immunosuppressed individuals^[10]. This variability in the prevalence rates and this association with diseases may be due to epidemiological differences, including geographic variability, morphological and genetic variations^[11]. The variations and the lack of standardization in diagnostic methods have led to controversy regarding its role as a human pathogen and its prevalence^[12,13].

Various *Blastocystis* subtypes have differences, such as pathogenicity, immune response, effects on microbiota, and drug resistance^[12,14,15]. Hence, there

are uncertainties regarding the clinical significance, pathogenic potential, and need of treatment for *Blastocystis* spp., especially in immunocompromised individuals^[16,17]. In addition to asymptomatic findings in blastocystosis symptomatic findings, such as acute or chronic diarrhea, abdominal cramps, nausea, and flatulence, are also seen^[18,19].

Bibliometrics is a widely used method for analyzing research topics, research status, and publication quality as objective measures^[20]. Using this method, meaningful information can be produced from large data and the issues that cause confusion can be clarified. For this purpose, this method has gained great interest in recent years and has started to become widespread in many different fields^[21]. As such, it has become an important issue to visualize the data obtained in bibliometric analysis studies, where complex relationships have been revealed in data mining, and to examine visualization approaches.

The purpose of this study was to evaluate potential future pathways for investigating *Blastocystis* spp. and to generate a comprehensive summary of the *Blastocystis* spp. literature. In this way, the issues that cause confusion about *Blastocystis* spp. can be clarified.

MATERIAL AND METHODS

This descriptive bibliometric study was conducted in the Çanakkale Onsekiz Mart University, Faculty of Medicine, Department of Infectious Disease and Clinical Microbiology, Çanakkale, Türkiye, during the period from January, 2023 to February, 2023.

Study design: Original articles published from 1st of January 1970 to 31st December 2022 were downloaded and analyzed using Web of Science (WoS) Core Collection (Clarivate Analytics, Philadelphia, USA).

Data collection: Total downloads included 1192 publications, of which 85 abstracts, 78 letters, 56 reviews, 35 notes, 18 proceeding papers, and 14 editorials were excluded. The study analyzed the remaining 906 original articles. Publication titles, authors, years of publication, countries, affiliations, keywords, publishing journals, abstracts, and citations were saved as text (TXT) files and imported into Microsoft Office Excel 2019 (Los Angeles, CA, USA).

Search strategy: The title of the search engine was examined using the selected keywords '*Blastocystis* spp.', or '*Blastocystis hominis*'. The time span selected was 1970-2022 extending from the 1st of January, 1970 to 31st December, 2022. The year 2023 was not included as it is not complete yet. Case reports, editorials, and letters were among the other publication types that were excluded from the search since they were not peer-reviewed publications.

Processing the WOS database outputs: Parameters used in WOS analysis included Hirsch (H) indices, number of citations, and international collaborations. In addition, the most scientific countries were compared among themselves.

Network analysis: The WoS database was used to retrieve the whole text data of the included publications. VOSviewer (version 1.6.10, Leiden University, Netherlands) was used to illustrate the collaboration network, emphasis, and future trends of the relevant topic in this bibliometric study. The data were imported using the VOSviewer software to analyze the citations, keywords, co-authorship, co-occurrence, and co-citation. Finally, three bubble maps (Figures 1-3) were generated to show the bibliometric analysis results. Colors in the bubble maps indicated the item clusters, while the distance and breadth of the lines between two bubbles reflected the frequency of co-occurrence. In the first map, the keywords that appeared at least 5 times are visualized in proportion to the number of articles in the selected time interval, and connection lines indicate how often the keywords are used together. This enabled us to see which keywords are mostly used in the studies. While the second map represents authors with at least 5 publications, and at least 100 references. The third map shows the international collaboration network conducted by country with connection lines indicating how frequently countries communicate with each other.

Ethical approval: The research conformed with the 2013 revision of the Helsinki Declaration. As there was no human or animal research, an ethics committee's permission was not necessary.

RESULTS

In the present descriptive bibliometric study, the following points were noted:

1. A total of 1192 documents were found, of which 906 were articles, and only those publications were analyzed. The first article on *Blastocystis* spp. was published in 1973, and it did not have any citations. Since 2000, there was a limited number of articles, and since 2014 there was a minimum of 34 publications per year. The highest number of articles was published in 2021 (n= 87) and the highest number of citations (n= 3,485) was in 2021 (Fig. 4). The articles were cited 24065 times in total and the mean Hirsch (H) index was 74. Most of them (95.58%) were published in English.
2. The USA (n: 93), Singapore (n: 79), China (n: 76), Malaysia (n: 74) and Japan (n: 59) were the countries that published the most articles on *Blastocystis* spp. Publications were recorded from 95 countries around the world, out of which Türkiye ranked 6th (Table 1). The table showed 25 out of 95 entries.

3. The articles were from 49 different research areas and mostly from Parasitology (n:413), Tropical Medicine (n:145), and Microbiology (n:143) research areas (Table 2). The table showed 25 out of 44 entries; 3 records (0.38%) did not contain data in the field being analyzed.
4. Authors from 1,017 different affiliations/universities contributed to the writing of the articles. The National University of Singapore was the leading affiliation on *Blastocystis* spp. research,

with 79 published articles (Table 3). The table showed 25 out of 873 entries 1 record (0.11%) did not contain data in the field being analyzed.

5. Most of the articles were published in the Journal of Parasitology Research (n: 144) (Table 4). The table showed 24 out of 268 entries.
6. Publishers (n: 130) that have published the most on *Blastocystis* are listed in table (5). The table showed 10 out of 130 entries.

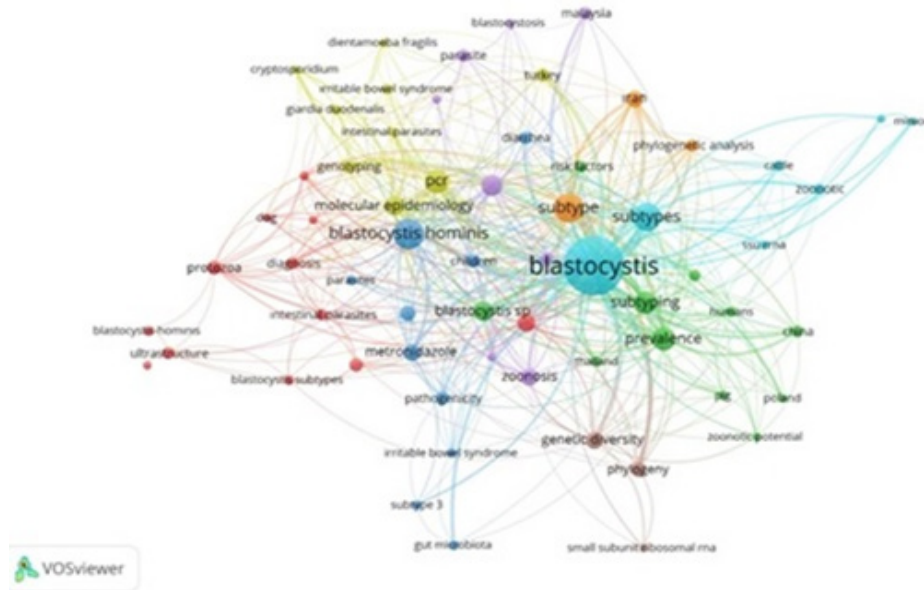


Fig. 1. Keyword visualization map of the articles with at least 5 occurrences. * Connecting lines are indicative of the occurrence relations in the articles. Keywords represented with a larger circle size or font size had a relatively higher occurrence in the articles.

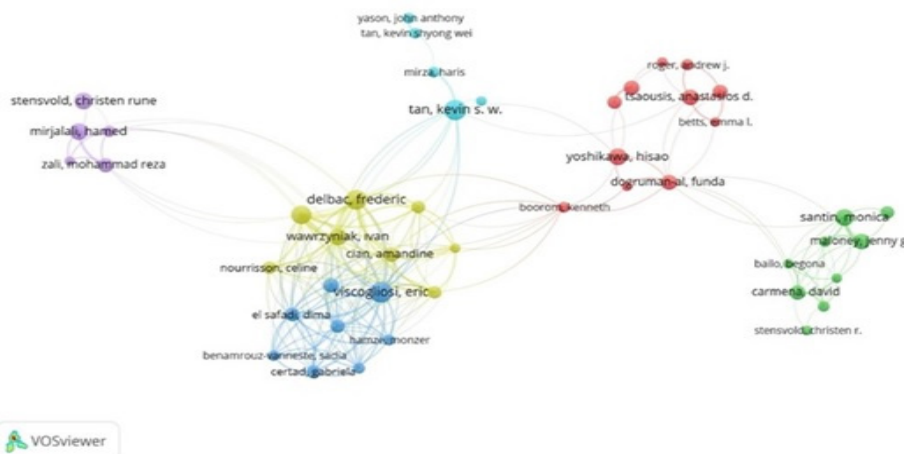


Fig. 2. Authors with at least 5 publications and 100 citations are shown on the map. **Citations are shown by lines linking authors. Authors with a greater circle size or font size had a higher number of citations.

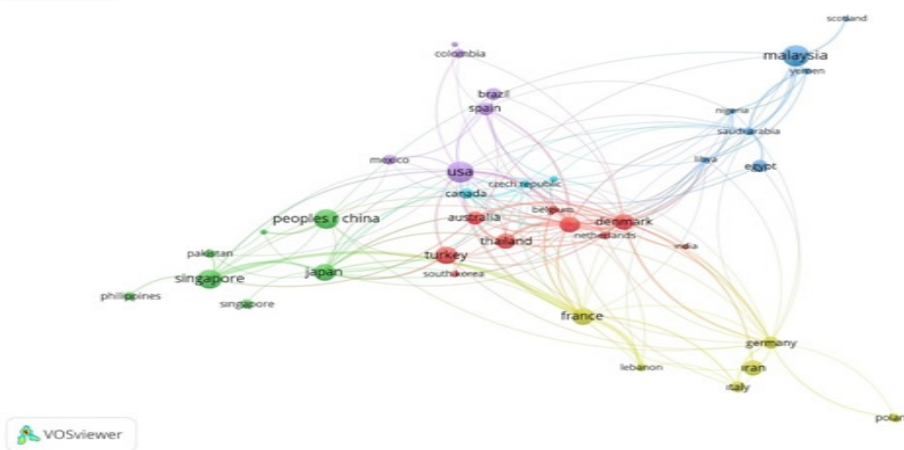
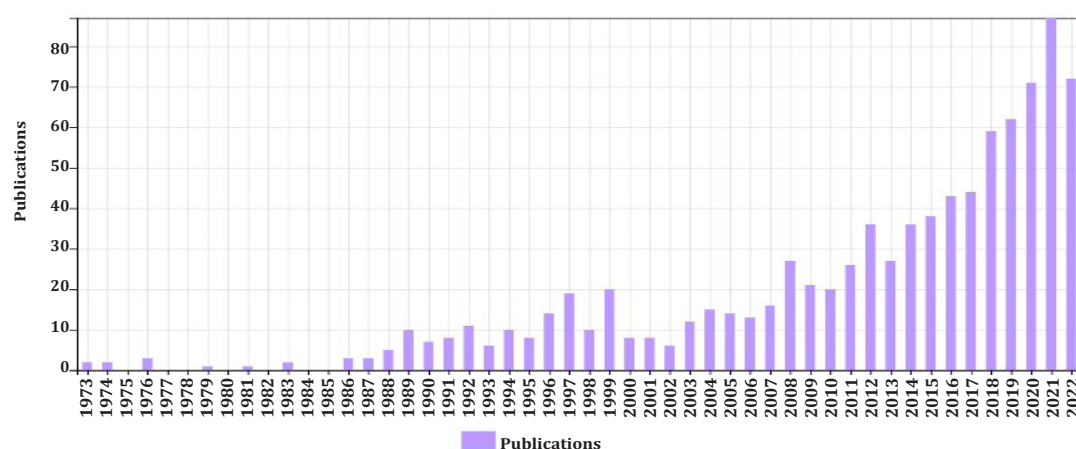


Fig. 3. International collaboration network map.

Table 1. Most productive countries on *Blastocystis* spp. research.

Country	No. of publications	% out of 906
The USA	93	10.26
Singapore	79	8.72
China	76	8.39
Malaysia	74	8.16
Japan	59	6.51
Turkey	58	6.40
Iran	55	6.07
England	50	5.52
France	50	5.52
Denmark	45	4.98
Australia	40	4.41
Thailand	40	4.41
Egypt	37	4.08
Spain	35	3.86
Brazil	29	3.20
Germany	28	3.09
Italy	23	2.54
Mexico	20	2.21
Pakistan	20	2.21
Canada	19	2.10
Colombia	19	2.10
Saudi Arabia	18	1.99
Poland	17	1.88
Czech Republic	14	1.55
Philippines	12	1.33

**Fig. 4.** Number of articles and citations between 1973 and 2022.**Table 2.** List of the articles according to research areas.

Research area	No. of publications	% out of 906
Parasitology	413	45.58
Tropical Medicine	145	16.00
Microbiology	143	15.78
Infectious Diseases	122	13.47
Public Environmental Occupational Health	87	9.60
Veterinary Sciences	70	7.73
Science Technology other Topics	45	4.97
General Internal Medicine	35	3.86
Immunology	29	3.20
Biochemistry Molecular Biology	28	3.09
Zoology	23	2.54
Gastroenterology Hepatology	21	2.32
Pharmacology Pharmacy	16	1.77
Environmental Sciences Ecology	14	1.55
Life Sciences Biomedicine Other Topics	10	1.10
Pediatrics	10	1.10
Biotechnology Applied Microbiology	9	0.99
Cell Biology	9	0.99
Research Experimental Medicine	9	0.99
Genetics Heredity	8	0.88
Agriculture	6	0.66
Medical Laboratory Technology	6	0.66
Pathology	6	0.66
Plant Sciences	6	0.66
Evolutionary Biology	5	0.55

Table 3. Most productive affiliations on *Blastocystis* spp. research.

Affiliations	No. of publications	% out of 906
National University of Singapore	79	8.72
Malaya University	60	6.62
Nara Women's University	42	4.64
Centre National de la Recherche Scientifique	41	4.53
Statens Serum Institut	40	4.42
Egyptian Knowledge Bank	36	3.97
Institut National de la Sante Et De La Recherche Medicale Inserm	34	3.75
Universite De Lille site	33	3.64
Universite De Lille	32	3.53
Institut Pasteur Lille	30	3.31
The International Network of Pasteur Institutes	30	3.31
Shahid Beheshti University Medical Sciences	24	2.65
The University of Clermont Auvergne	24	2.65
Institute of Biological Sciences	23	2.54
Institute of Ecology and Environment	20	2.21
The University of London	19	2.10
The Aga Khan University	18	1.99
The United States Department of Agriculture	18	1.99
Ain Shams University	17	1.88
The London School of Hygiene & Tropical Medicine	17	1.88
Gazi University	16	1.77
Centre Hospitalier Universitaire de Lille	15	1.66
The National Institutes of Health	15	1.66
Gifu University	14	1.55
El Instituto de Salud Carlos III	14	1.55

Table 4. Journals that published the most articles on *Blastocystis* spp. research.

Journal titles	No. of publications	% out of 906
Parasitology Research	79	8.72
Parasites Vectors	60	6.62
Plos one	42	4.64
American Journal Of Tropical Medicine and Hygiene	41	4.53
Parasitology International	40	4.42
Infection Genetics and Evolution	36	3.97
Parasitology	34	3.75
Acta Tropica	33	3.64
Veterinary parasitology	32	3.53
International Journal for Parasitology	30	3.31
Revista da Sociedade Brasileira de Medicina Tropical/Journal of the Brazilian Society of Tropical Medicine	30	3.31
Microorganisms	24	2.65
Experimental parasitology	23	2.54
Journal of Clinical Microbiology	20	2.21
Acta Parasitologica	19	2.10
European Journal of Clinical Microbiology Infectious Diseases	18	1.99
Parasite Epidemiology and Control	18	1.99
Tropical Biomedicine	17	1.88
Iranian Journal of Parasitology	17	1.88
Journal of Eukaryotic Microbiology	16	1.77
Korean Journal of Parasitology	15	1.66
Scientific Reports	15	1.66
Comparative Immunology Microbiology and Infectious Diseases	14	1.55
The Southeast Asian Journal of Tropical Medicine and Public Health	14	1.55

Table 5. Publishers with the most publications on *Blastocystis* spp.

Publishers	No. of publications	% out of 906
Springer Nature	250	27.59
Elsevier	186	20.53
Mdpi	37	4.08
Public Library Science	34	3.75
Wiley	30	3.31
Amer Soc Microbiology	27	2.98
Amer Soc Trop Med & Hygiene	22	2.43
Cambridge Univ. Press	21	2.32
Oxford Univ. Press	19	2.10
Frontiers Media Sa	13	1.44

DISCUSSION

Blastocystis spp. was first identified in 1911 and named as a fungus. After *Blastocystis* spp. was accepted as a non-pathogenic agent in a study, it was not emphasized much and not many studies were conducted on it. However, after Zierdt *et al.* stated in a study in 1967 that the physiological and morphological structure of *Blastocystis* spp. showed a protozoan character, that it could be a pathogenic agent, and that it had different forms, studies and publications on this agent began to increase^[22]. Later, Zierdt examined its morphological and physiological features and classified *B. hominis* as a protozoan^[23]. Later it was taxonomically classified in 1996^[24] and its pathogenicity has been emphasized since. Now, a new aspect of *Blastocystis* spp. is introduced every day and related studies are increasing day by day. Recently with the application of modern phylogenetic techniques such as the search for 18s rRNA, its taxonomic classification was made in recent years, placed it in the Stramenopile kingdom^[24].

Blastocystis spp. is an oral-fecal transmitted agent. The most important reason for the transmission of the disease is the lack of attention to hygiene conditions. However, being in close contact with animals, the geographical region and climatic conditions also affect the transmission and prevalence of the disease. When the epidemiology of the disease is considered, it appears to be at high rates ranging from 30% to 50% in developing countries. In developed countries, this rate is between 1.5% and 10%^[19]. The most important point in the evaluation of the epidemiological data of *Blastocystis* spp. is to determine the distribution at the subtype level. Because *Blastocystis* spp. has pathogenic and non-pathogenic subtypes, investigating the prevalence of these subtypes is more meaningful in determining the risks of that region. For this reason, researchers mostly focus on the diagnosis and distribution of the subtypes^[19]. Different results have emerged in epidemiological studies conducted around the world.

The lowest rate is seen in Japan, at 0.5-1% and Singapore, at 3.3%. In some countries, different rates have been reported in different studies, such as Argentina at 27.2%, Brazil at 40.9%, Indonesia at 60%, Egypt at 33.3%, China at 1.9-32.6%, and Thailand at 0.19-45.2%^[25]. The distribution at the subtype level around the world is as follows: Subtype-3 (ST3) is seen as the most dominant subtype in many countries around the world, such as Thailand at 41.7-92.3%, Egypt at 54.6%, Singapore at 78%, Germany at 21%, and France at 53%^[25]. In addition, different subtypes were seen at a higher rate in a limited number of studies. Subtype-4 (ST4) was seen in 94.1% in Spain, 63% in France, and 84% in Nepal. Subtype-1 was found at a rate of 51.4% in China and 41% in Brazil when compared to the other subtypes^[26]. In fact, our study confirms these

data, because every country has its own unique social structure. The number of immunosuppressed individuals in these populations can change the visibility of blastocystosis. In addition, different subtypes can be seen in societies dependent on animal husbandry. In addition, in societies with insufficient health infrastructure, the identification of this parasite may not be possible because laboratory facilities are insufficient.

Blastocystis spp. is an extracellular agent that lives in the gastrointestinal tract and is very common in the community. This agent is discussed in many aspects today. One of the most discussed issues is whether *Blastocystis* spp. is pathogenic. *Blastocystis* spp. can be found without any symptoms in infected individuals, or it can cause nonspecific infections that lead to simple enteric complaints. It may also play a role in more specific diseases, such as irritable bowel syndrome and urticaria^[27].

The clinical significance of *Blastocystis* spp. is controversial^[28], however, as a result of data obtained from multidisciplinary studies and findings in *in vitro* and *in vivo* studies, the view that *Blastocystis* spp. is a pathogenic agent is more accepted. The treatment of individuals infected with *Blastocystis* spp. is controversial because of the indeterminate pathogenesis of the parasite and the self-limiting nature of the disease. Treatment is recommended when the infection has a chronic course and no other factor can be detected^[29,30]. It is important to use advanced molecular diagnostic techniques to eliminate the uncertainties regarding the pathogenicity, immune response, effect on the microbiota, prevalence and/or drug resistance of *Blastocystis* spp.^[31]. Providing clean water and sanitation for all, better hygiene conditions, and reliable waste management are essential for the prevention and protection of human health against *Blastocystis* spp.

The prevalence of intestinal parasitic infections in immunocompromised individuals in developing countries reached up to 95%. Protozoa and helminths cause these infections and their primary clinical finding is diarrhea^[32]. *Blastocystis* spp. is one of the common intestinal protozoa to infect humans^[17]. While records on *Blastocystis* spp. are mostly not archived in developing countries, different diagnostic techniques are employed in these countries. Therefore, the prevalence of *Blastocystis* spp. may vary in the literature^[11,12,33]. The uncertainty of the data about the prevalence of *Blastocystis* spp. in developed and developing countries, the fact that this data may vary according to the geographical regions, and the diagnostic method used prompted us to conduct this research.

Researchers perform bibliometric analysis for a variety of reasons, including investigating the uncertainty of a particular field in previous studies, as well as determining collaborations and study topics. Bibliometric analysis is a method that processes quantitative data^[34], and has been used seriously in the field of medicine in recent years^[34-39]. Data obtained by bibliometric analysis are often large and objective (citations, publications, keywords, and topics), but its interpretations are often based on both objective (performance analysis) and subjective (thematic analysis) assessments generated through informed techniques and procedures. Bibliometric analysis is a very effective method to easily interpret complex issues and reach a meaningful result by processing large data^[37,38]. As a result, a well-performed bibliometric analysis provides a better understanding of complex or ambiguous topics. The fact that there are subjects in need of explanation in the field of parasitology has also encouraged scientists in this field to obtain meaningful information from publications^[35,37,40-42]. Therefore, due to the increase of reported cases of *Blastocystis* spp. in recent years and the increase in uncertainties on this subject, a bibliometric research is needed.

It should be stated that databases such as Scopus and Wos have facilitated the acquisition of large volumes of bibliometric data. Bibliometric software, such as Gephi, Leximancer, and VOSviewer, allow these data to be processed and made meaningful. In our study, all of the data and analyses were conducted in English, and the Wos database was used for the bibliometric data and the VOSviewer program was used for the visualizations.

Blastocystosis has attracted the attention of scientists working in the fields of microbiology and infectious diseases, especially in Parasitology. The parasite received an increasing interest all over the world, especially in the USA. Globally out of the 95 countries that published articles on *Blastocystis* spp. research, the leading countries were the USA (n = 93; 10.265%), followed by Singapore (n = 79; 8.720%), and China (n = 76; 8.389) (Table 1). The detection and prevalence of *Blastocystis* spp. has increased due to the use of advanced diagnostic techniques, the detection of subtypes associated with pathogenicity^[43,44], the increase in close contact with animals^[45] and the increase in immunocompromised individuals^[46]. So there has been a significant increase in the number of articles and citations especially since 2000.

Out of the total 906 publications from different research areas, 413 (45.58%) were from Parasitology, 145 (16.00%) from Tropical Medicine, and 143 (15.78%) were from Microbiology (Tables 1 and 2). With the increasing interest in *Blastocystis*

spp. in the world, there has also been an increase in the acceptance and publication of the studies conducted in this field by publishers (Tables 4 and 5). Bibliometric studies help to both reveal the current status of a scientific subject and to shape future scientific research. It is believed that this study will be useful for scientists who want to do more research on this subject. The USA, Singapore, and China have led scientific production research on *Blastocystis* spp. Efforts should be made to help them to develop interdisciplinary scientific research networks on *Blastocystis* spp.

Finally, our study had a few limitations, including the inability to examine and analyze papers from publications that were not indexed in the WOS indices. Additionally, since the keywords were only in English, articles in other languages may have been excluded. Other databases, such as Scopus, were not included in the study and only VOS viewer was used to conduct the analyses. Furthermore, content analysis was not performed, and more detailed comparative analyses may be planned for future studies.

Author contribution: Bilden A and Ekici A contributed in conceptualization, methodology, data curation, and writing- original draft preparation. Çiçek M, and Aydemir S performed visualization, investigation, supervision, software, and validation. Gürbüz E, Ünlü AH, and Alkan S wrote, reviewed and edited the manuscript.

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