

Original research on Behçet's syndrome: a bibliometric analysis over 20 years (2000-2019)

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Abstract Objective

To perform a bibliometric analysis of original research articles on Behçet's syndrome (BS) published over the last 20 years prior to the COVID-19 pandemic, and to systematically describe their characteristics and citation records.

Methods

PubMed database was searched for any article published on BS between 2000 and 2019. We identified all original research articles and categorised them by country of origin and type of research, i.e., clinical, translational and basic. Each article's impact was assessed using the individual citation numbers from Google Scholar search engine; we also calculated the median annual citation rates (ACRs), both per country and research type.

Results

Of a total of 2,381 retrieved original articles from 51 countries, the majority reported on clinical (52.6%), followed by translational (46.0%) and basic research (1.4%). Turkey had the highest number of publications (39% of articles) followed by four countries (Korea, China, Japan, Italy) where BS is also relatively prevalent. However, regarding median ACRs, France was first, followed by United Kingdom, Germany and Collaboration. Although the number of articles has almost doubled between 2010-2019 versus 2000-2009, median ACRs across either clinical or translational research had a downward trend.

Conclusion

Researchers from countries where BS is prevalent are more productive, albeit their work is of lower impact compared to countries with generally higher research budgets. A considerable increase of original research articles on BS is observed over time but further funding may be warranted for a parallel increase in the respective scientific impact.

Key words

Behçet's syndrome, bibliometrics, citations, country, research

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Introduction

Adamantiades-Behçet's or Behçet's syndrome (BS) is a chronic inflammatory and systemic vasculitic disorder classified in the group of variable vessel vasculitis (1). It is characterised by recurrent oral and genital aphthous ulceration, uveitis, skin lesions, arthritis and cardiac, pulmonary, gastrointestinal, neurological and vascular involvement (2). The epidemiology of BS is unique, being distributed worldwide but it is more common in countries along the Ancient "Silk Route", extending from Japan to the Middle Eastern and the Mediterranean countries (3). Turkey has the highest prevalence (BS affecting up to 400 out of 100,000 population) followed by other Middle Eastern countries. However, the prevalence of BS is thought to be gradually increasing in many countries worldwide due to immigration trends and due to its better understanding and diagnosis.

The geographic distribution of human leucocyte antigen (HLA)-B51 allele in the Ancient "Silk Route", along with environmental factors and possible infectious agents, are believed to trigger BS emergence. Its pathogenesis lies at the intersection between autoimmune and autoinflammatory mechanisms, with T-cell homeostasis perturbations and neutrophil migration playing a central role (4-6).

Since its first description, an increasing body of clinical and translational research has been published. However, little research has been conducted to examine publication trends in the BS literature (7). Bibliometric analysis involves the application of mathematical and statistical methods to scientific publications (8) in order to assess group's research activity, performance and scientific quality. Bibliometric reports present a snapshot of the global literature on a certain topic (9), including information on the productivity of the countries and authors, total number and impact of published articles and their distribution by country or by language (10). In light of this, publishers and authors could determine more efficiently the manuscripts critical to patient care improvement, scientists may focus their future research on certain ar-

reas, and possible funds can be assigned accordingly (11).

Based on the above, it seems valuable to examine publication trends of original research in a quantitative and qualitative perspective and understand impact of research in BS over a long period of time. The bibliometric method is valuable in this pursuit, as it has been used in various scientific fields, including rheumatology (12-14). Therefore, the aim of this study was to perform a quantitative and qualitative analysis of original research articles on BS that were published over the last 20 years prior to the COVID-19 pandemic using bibliometric methods.

Materials and methods

PubMed database was searched for any article published on BS between January 1, 2000, and December 31, 2019. Literature reviews, systematic reviews, meta-analyses, case reports, guideline statements, editorials, comments, historical articles and errata were excluded. The original research articles in English or articles with available abstracts in English with the term "Behçet" or "Behçet's" in their title, were eligible for inclusion in the study. After excluding duplicate papers, all included articles were subsequently categorised as clinical, translational or basic, depending on the type of original research.

Descriptive statistics were used to calculate the number of articles in total and by research type throughout the literature review period, as well as to assess the contribution of each country in a quantitative and qualitative perspective. The citation number of each paper was obtained from the Google Scholar search engine. The term "Citation score" or "Annual citation rate" (ACR) is defined as the total number of the citations each article acquired up to 2020 to the number of years (2020 - year of publication). This term is used to evaluate and compare the impact of the published original research articles, irrespective of the differences in the year of publication. It is also adopted in order to list the top 100 cited articles based on their country of origin. Median ACR is the term used for the median of the ACRs of the articles and

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its distribution is analysed per year and per study type. We used STATA v. 13.0, College Station, Texas, USA for all the statistical analyses.

The country of origin of each publication is based on the first author information, as this is noted in each article. The term "Collaboration" is used when there are more than one country participating in the production of the article or when the author of the article is an international team.

Results

The PubMed literature search resulted in 2,381 research articles for analysis, 1,252 (52.6%) of which were classified as clinical, 1,095 (46.0%) as translational, and 34 (1.4%) as basic research articles.

In Table I, the contribution of each country in BS publications is presented in total and per study type between 2000 and 2019. Turkey is the country with the most publications (921 articles) followed by Korea (253 articles), China and Japan. Additionally, Turkey had the highest number of articles concerning the clinical (513 articles) and translational (405 articles) research publications, once again followed by Korea, Japan and China. However, Korea had the highest number of publications on basic research, publishing more than half of the total number of basic research articles between 2000 and 2019.

Regarding the number of citations by country and study type, Turkey had 24,171 citations in all study types, which is more than one third of the total citation number of all the countries included in our study (60,997 citations in total). Korea and Japan followed with 6,686 and 5,507 total citation numbers, respectively. A similar trend applies when it comes to clinical and translational studies separately. In basic studies, Korea had the highest number of citations (288 citations) (Table I).

In general, the total number of original research articles on BS have increased throughout the literature review period, ranging from 59 in 2000 to 193 in 2019 (Fig. 1a). This trend is also reflected in the increase of the clinical and translational research publications between 2000 and 2019. In specific, there are 34

Table I. Total number of original articles 2000-2019 per country and per study type/in total. (In the brackets there is the citation number of the aforementioned articles per country and per study type/in total) *

Countries (in alphabetical order)	Clinical studies	Translational studies	Basic studies	Total number of studies
Algeria	3 (3)	13 (161)	1 (9)	17 (173)
Argentina	0	1 (0)	0	1 (0)
Armenia	0	1 (11)	0	1 (11)
Australia	0	1 (6)	0	1 (6)
Austria	1 (2)	1 (13)	0	2 (15)
Brazil	16 (268)	6 (72)	0	22 (340)
Canada	2 (15)	0	0	2 (15)
Chile	1 (8)	1 (14)	0	2 (22)
China	74 (712)	116 (2260)	1 (6)	191 (2978)
Collaboration	12 (386)	7 (533)	0	19 (919)
Colombia	1 (0)	0	0	1 (0)
Denmark	1 (6)	0	0	1 (6)
Egypt	24 (139)	25 (227)	0	49 (366)
France	32 (1428)	9 (524)	0	41 (1952)
Germany	18 (1014)	12 (455)	0	30 (1469)
Greece	14 (1143)	3 (44)	0	17 (1187)
India	3 (46)	0	0	3 (46)
Iran	54 (1248)	50 (439)	1 (10)	105 (1697)
Iraq	7 (390)	3 (41)	0	10 (431)
Ireland	0	1 (2)	0	1 (2)
Israel	16 (603)	16 (729)	0	32 (1332)
Italy	59 (1465)	49 (976)	1 (9)	109 (2450)
Japan	93 (2746)	92 (2761)	0	185 (5507)
Jordan	2 (7)	0	1 (6)	3 (13)
Korea	123 (3671)	111 (2727)	19 (288)	253 (6686)
Lebanon	3 (77)	4 (35)	0	7 (112)
Mexico	0	1 (25)	0	1 (25)
Morocco	15 (159)	4 (113)	0	19 (272)
Netherlands	5 (127)	7 (174)	0	12 (301)
New Zealand	1 (0)	0	0	1 (0)
Nigeria	1 (6)	0	0	1 (6)
North Cyprus	1 (20)	0	0	1 (20)
Pakistan	0	0	1 (0)	1 (0)
Poland	1 (3)	0	0	1 (3)
Portugal	4 (39)	11 (182)	0	15 (221)
Romania	2 (21)	0	0	2 (21)
Russia	8 (29)	2 (8)	0	10 (37)
Saudi Arabia	9 (340)	2 (7)	0	11 (347)
Senegal	3 (18)	0	0	3 (18)
Serbia	3 (39)	1 (11)	0	4 (50)
Spain	27 (551)	30 (697)	4 (45)	61 (1293)
Sultanate of Oman	3 (46)	0	0	3 (46)
Sweden	1 (52)	0	0	1 (52)
Switzerland	3 (139)	2 (38)	0	5 (177)
Syria	0	1 (11)	0	1 (11)
Taiwan	13 (166)	8 (128)	0	21 (294)
Thailand	4 (18)	1 (7)	1 (10)	6 (35)
Tunisia	25 (548)	54 (1548)	0	79 (2096)
Turkey	513 (14357)	405 (9809)	3 (5)	921 (24171)
UK	29 (1278)	27 (1123)	1 (50)	57 (2451)
USA	22 (729)	17 (586)	0	39 (1315)
Total	1252 (34062)	1095 (26497)	34 (438)	2381 (60997)

*UK: United Kingdom, USA: United States of America.

clinical research articles published in 2000 and 128 in 2019. In 2000, there are 25 published translational research articles, and this number has increased to 61 in 2019. However, the production of basic research publications is low when compared to the other study

types, as they are only 34 in total (Fig. 1b).

In order to assess the impact of the published research, we have calculated the ACR of each article, the definition of which is analysed in the Methods section. Based on this, we have also cal-

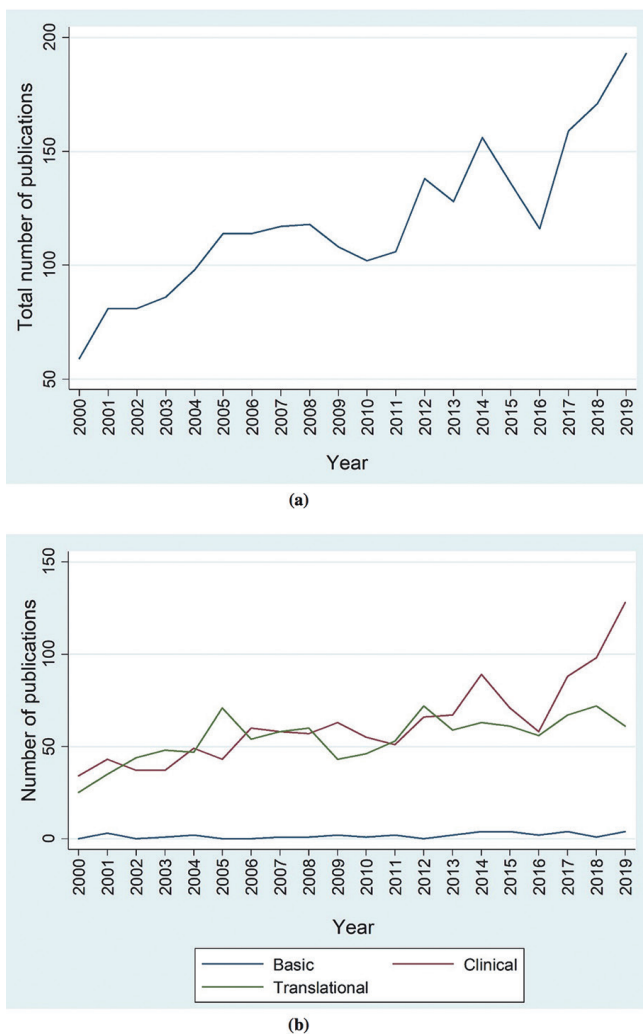


Fig. 1. Number of original articles 2000-2019 per year. (a) total; (b) per research type (basic, clinical, translational).



Fig. 2. Median annual citation rate (ACR) of original articles published between 2000-2019 per year. (a) total; (b) per research type (basic, clinical, translational).

culated the median ACR of all publications per year in total and per research type, as described in Figure 2.

In general, the total median ACR for the articles published between 2000 and 2019 is approximately 1.56, showing a downwards trend in the figure (Fig. 2a). The median ACR of clinical, translational and basic research publications for the same period is 1.56, 1.57 and 1.13, respectively. For the clinical research articles, the median ACR has two peaks; in 2003 and 2010. As far as clinical and translational research articles are concerned, a decreasing trend between 2000 and 2019 is observed. The basic research graph is difficult to interpret, as there are zero or only one publications in many of the years included in the literature review period (Fig. 2b).

Table II summarises the impact of the published research by country of origin. The countries are ranked based on their total citation number and the top 20 countries are listed. In this table, we have calculated the median ACR of each of these 20 countries, in total and per study type. Out of the top 20 countries with the highest total citation number, France is the one with the highest total median ACR going up to 4.00, followed by United Kingdom (UK) (with a median ACR around 3.00), Germany and Collaboration. For reasons of comparison, Turkey's median ACR is 1.47. When it comes to clinical studies, France has the highest median ACR (approximately 4.00) followed by Germany, UK and Israel. As far as translational studies are concerned, UK

is in the first place, with a median ACR of 3.33, followed by Collaboration, United States of America (USA) and Israel. In addition, UK, with only one basic study, has the highest ACR when it comes to basic research, with Spain and Italy in the second and third place, respectively.

The top 100 cited articles, ranked according to their ACR, by country of origin are presented in Table III. Information (title, authors, year of publication, type of research, country of origin) of the top 10 cited articles (ranked by their ACR) is included in Supplementary Table S1. Turkey is the country with the highest number of original research articles that are in the top 100 when it comes to their ACR, as there are 27 Turkish publications in Table III, with

Table II. Median annual citation rate of original articles (clinical, translational, basic studies and in total) published between 2000-2019 by country (country ranking according to total citation numbers; only the top 20 countries are listed).

Countries (ranking by total citations)	Median annual citation rate			
	Clinical studies	Translational studies	Basic studies	Total
Turkey	1.45	1.50	0.50	1.47
Korea	2.00	1.67	1.00	1.73
Japan	1.83	1.96	-	1.89
China	1.00	1.93	1.00	1.25
UK	2.67	3.33	10.00 (1 study)	3.00
Italy	2.12	1.45	1.80	2.00
Tunisia	1.21	1.92	-	1.82
France	4.00	2.20	-	4.00
Iran	1.82	0.83	1.67	1.33
Germany	3.57	2.34	-	2.75
Israel	2.60	2.53	-	2.60
USA	2.04	2.56	-	2.25
Spain	2.00	1.73	3.33	2.00
Greece	1.61	0.56	-	1.60
Collaboration	1.93	2.67	-	2.67
Iraq	2.27	0.76	-	1.73
Egypt	0.71	1.34	-	1.14
Saudi Arabia	2.00	0.73	-	1.45
Brazil	1.95	0.82	-	1.77
Netherlands	0.88	1.64	-	1.32

UK: United Kingdom, USA: United States of America.

5 of them in the top 10. Turkey is followed by Japan, Italy and France, with 12, 9 and 9 articles in the top 100 cited articles, respectively. The ACR range of the top 100 cited articles is between 48.10 and 8.31.

Discussion

In a quantitative perspective, our results show that there is an increasing trend in the number of original research articles on BS between 2000 and 2019. Our decision to exclude 2020 and 2021 was based on the fact that publication of original research articles in any field was destabilised during the COVID-19 era for many obvious reasons, including perhaps some slackness of critical

peer reviewing. Notably, the number of research articles has almost doubled between 2010-2019 *versus* 2000-2009, indicating an ongoing scientific research development in the BS field. This finding is consistent with those in other bibliometric analyses (7,9) which report a considerable growth of BS literature between 1990 and 2010, as well as between 2010 and 2019, respectively. In the first analysis, Shahram *et al.* (7) used the Web of Science database to describe publication trends, geographical distribution and citation profile of all publications relevant to BS between 1990 and 2010, in any language. Using also the Web of Science database, Kocyigit and Akyol (9) performed a

second bibliometric analysis of original and review articles on BS published in English only in the period 2010-2019, assessing their numbers, country of origin, citation and altmetric data.

A strength of our study is that all original articles on BS published in PubMed indexed journals were categorised to clinical, translational and basic research articles, depending on the type of the published research. The majority of the included articles consists of clinical research, followed by translational research. However, one point for discussion is the persistently low production of basic research publications, especially given the fact that BS pathophysiology is characterised by an interrelationship of genetic and environmental factors that are unknown (2-5).

As far as geographic distribution is concerned, an association is observed between prevalence of the disease and a country's contribution to BS literature. In light of this, Turkey, which has the highest registered prevalence, has also by far the highest productivity, followed by other countries where BS is prevalent, such as Korea, China and Japan. Our results agree to a great extent with those reported by Kocyigit and Akyol (9) while Shahram *et al.* (7) and Şenel *et al.* (10) note that, in their bibliometric analyses, Turkey, Japan and USA are the top three contributors in BS literature. In our study, roughly 39% of papers come from Turkey and 31% of papers come from four other countries, meaning that 70% of publications come from 5 out of 51 publishing countries (including Collaboration), across all continents, that are included. Shahram *et al.* (7) also mention that, as in our study, there are countries with

Table III. Top 100 cited original articles published between 2000 and 2019 ranked by annual citation rate per country (countries are listed in alphabetical order).

RANK (annual citation rate range)	China	Collaboration	France	Germany	Greece	Iran	Italy	Japan	Korea	Netherlands	Saudi Arabia	Spain	Tunisia	Turkey	UK	USA
1 - 20 (48.10 -16.25)	1	2	3	0	1	0	0	1	0	0	0	0	1	10	0	1
21 -40 (16.20 -12.50)	1	0	2	2	1	2	2	2	0	0	0	0	0	8	0	0
41 -60 (12.43-10.77)	1	0	3	0	0	1	3	3	5	0	0	1	0	2	0	1
61 -80 (10.63-9.50)	2	0	1	0	0	0	3	2	1	0	1	2	1	4	2	1
81-100 (9.43- 8.31)	2	0	0	0	1	1	1	4	2	1	0	0	2	3	2	1
Total	7	2	9	2	3	4	9	12	8	1	1	3	4	27	4	4

UK: United Kingdom, USA: United States of America.

high incidence and prevalence of BS but lower contributions, like Spain, Saudi Arabia and Israel.

In a qualitative perspective, our results show that the quality and impact of the original research articles may be decreased over time, as the median ACR across either clinical or translational research has a descending trend. When evaluating the impact of the original articles published between 2000 and 2019 in relation to their country of origin, an association is observed between the level of production (quantity) and the citation number (quality) of the publications in total. This finding coincides with those reported in previous bibliometric analyses. (7, 9) For instance, Turkey has the highest citation number (24,171 citations) in our study, followed by Korea and Japan. All these countries have published great numbers of original research articles. However, there are countries with low production but with a relatively high citation number, such as UK, France and Greece.

One point for discussion is that the total citation number is not necessarily in association with the median ACR of the countries of origin. This finding is also applicable when it comes to study types separately. For example, even though Turkey, Korea and Japan are the top three countries in total citation numbers, their median ACR is lower than other countries with lower citation numbers, such as France, UK, Germany and Israel. In fact, France with a total number of 1952 citations, has the highest median ACR in total. A similar observation is made by Kocyigit and Akyol (9) as their results show that France, USA and UK are the top three countries, regarding the average citation value per article. Interpretation of these findings may take into account the fact that the aforementioned countries are characterised by bigger research budgets and a general scientific growth, leading per-

haps to higher impact articles than other countries where BS is more prevalent.

Lastly, our study shows that Turkey, which has the highest productivity, total citation number and prevalence of BS, also has the biggest number of publications in the top 100 cited articles, compared to other countries. More than one fourth of the 100 articles are of Turkish origin, and 5 of them are in the top 10. Moreover, the top cited article is collaborative, followed by another collaborative article and then by a Turkish one. The majority of the top 100 cited articles include clinical research, even though the production of clinical and translational research articles between 2000 and 2019 does not differ greatly.

To conclude, this is the first bibliometric analysis of BS to categorise the original articles by type of published research, in a large literature base spanning 20 years for analysis. It explores in a systematic way the quantitative and qualitative features of original research publications and it shows the growing scientific interest in BS, as reflected by the progressive growth in the number of articles during the pre-pandemic years. It also assesses the impact of the published scientific work, and includes important information about different countries' contribution in BS literature. Taken together, our findings suggest that further funding is warranted in BS research field, for a parallel increase in the respective scientific impact.

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