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ORIGINAL ARTICLE

Strengthening the Concept of Burden in Congenital Heart Diseases as a Contributor in Achieving Sustainable Development Goals: A Bibliometric Study

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Abstract

Introduction: Since more children with congenital heart disease (CHD) are surviving, policy changes are needed to improve their survival and quality of life. Global data and a literature review were systematically analyzed using The Global, Regional, and National Burden of CHDs, Global Burden of Disease (GBD) Study, published in *The Lancet Child & Adolescent Health*, which provided estimates of mortality from CHD.

Objective: To outline studies with CHDs and the concept of burden as a possible contributor to the achievement of sustainable development goals (SDGs) over time.

Methods: Bibliographic references and related authors (2005 to 2022) were researched and analyzed. Web of Science (WoS), Clarivate Analytics, was chosen as the source database to outline the results through bibliometric analysis, which is characterized by a quantitative and statistical technique to understand and measure new issues, as well as identify some trends in current research, regarding the issue of burden in CHDs.

Results: Sixty-five selected publications were found according to the bibliographic survey using the keywords "Burden CHDs." Afterwards, bibliometric analysis was performed, filtering the following results: years of publication; countries/regions; authors and number of citations; citation analysis; WoS index; types of documents; and research areas. The results are presented as figures.

Conclusion: The concept of burden addressing the issue of CHD has been used with great propriety for the development of goals for care of CHDs, mainly in the last decade, highlighting the year 2020.

Keywords: Global Burden of Disease; Congenital Heart Defects; Chronology.

Introduction

About the Global Burden of Disease (GBD) Study 2017

According to *The Lancet Child & Adolescent Health 2020*, published online in March 2020, the GBD Study is the most comprehensive worldwide observational epidemiological study to date. It describes mortality and morbidity (the rate of incidence of a disease) from major diseases, injuries, and risk factors to health at global, national, and regional levels. Examining trends from 1990 to the present and making comparisons across populations could enable the understanding of changing health challenges facing people across the world, in the twenty-first century. Published in October 2020, *The Lancet*'s special issue on GBD includes the most up-to-date global health data from 2019, with the latest analysis focused on 5 key themes: demographics, diseases and injuries, risk factors, population forecasting, and universal health coverage. By tracking progress within and between countries, GBD provides an important tool to inform clinicians, researchers, and policymakers, promote accountability, and improve lives worldwide.[1]

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Central Illustration: Strengthening the Concept of Burden in Congenital Heart Diseases as a Contributor in Achieving Sustainable Development Goals: A Bibliometric Study

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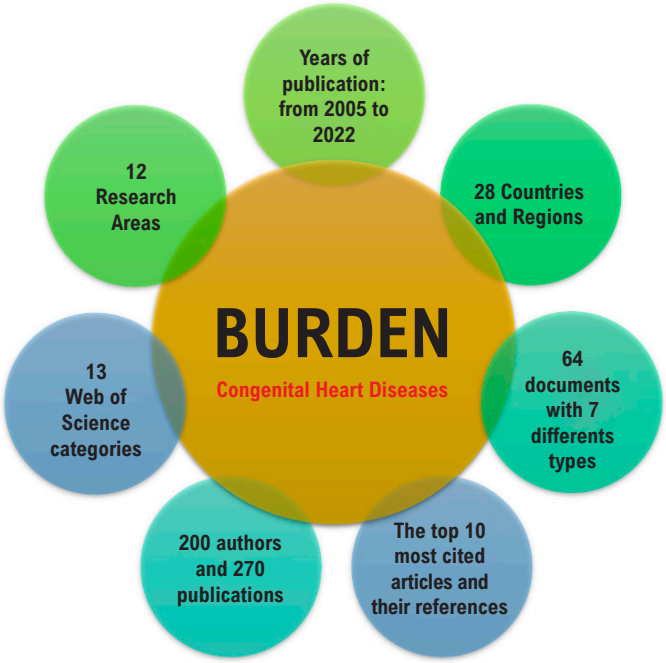

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Global, regional, and national burden of congenital heart disease (CHD), 1990 to 2017: a systematic analysis for the GBD Study 2017

This study is a systematic analysis of the GBD Study 2017, completed as part of the GBDs, Injuries, and Risk Factors Study 2017, which aimed to provide comprehensive estimates of CHD mortality, prevalence, and disability by age for 195 countries and territories from 1990 to 2017. It also provided important epidemiological data, including trends over time, on contributions to global morbidity and mortality. The GBD 2017 CHD Collaborators' study has provided new information on the burden of CHD throughout the world and presented important new epidemiological data not available in previous GBD analyses.[2] According to Bistra Zheleva, Vice President of Global Strategy and Advocacy of Children's Heart Link and co-author of the study, it was the first-ever Global Burden of CHD study published in *The Lancet Child & Adolescent Health*, in January 2020. As she pointed out, "The study is truly a milestone in the global health field. It provides new and important data to support our global advocacy efforts to help more children".[3]

The concept of burden

Ultimately, the concept of burden has properly been used in global theme issues related to diseases worldwide. The word "burden," as a noun, is defined by the Macmillan American English and British English Dictionary (2009 to 2020) as "a serious or difficult responsibility that one has to deal with; the responsibility of paying an amount of money, especially, when this is considered too much; a negative feeling that is difficult to deal with and that you cannot get rid of; something heavy that you have to carry." Adjectives frequently used with burden include: considerable, enormous, and heavy.[4] According to Collins Dictionary, burden, as countable noun, implies that, "If you describe a problem or a responsibility as a burden, you mean that it causes someone a lot of difficulty, worry, or hard work; A burden is a heavy load that is difficult to carry" [formal][5]

The use of the word "burden" as described above in both publications (Global, Regional, and National Burden of CHD 1990-2017 as part of the GBD Study 2017) has indicated the comprehensiveness of the term as a concern, that is, a robust pillar of sustainability of diseases , including CHD, worldwide.

CHDs: an overview

CHD, a major cause of serious morbidity and mortality, is common. It is usually defined as clinically significant structural heart disease present at birth. Although the incidence of CHD is similar worldwide, the burden of supporting these patients falls more heavily on countries with high fertility rates. The incidence of CHD at birth (sometimes referred to as birth prevalence) depends on how a population is studied. Before the introduction of echocardiography, incidence figures ranged from 5 to 8 per 1000 live births, but improved diagnosis has detected many more cases with milder forms, so that current estimates range from 8 to 12 per 1000 live births. Much depends on how early and how intensively diagnosis is made.[6]

According to the interpretation of the systematic analysis for the GBD Study 2017, CHD is a large, rapidly emerging global problem in child health. It accounts for nearly one third of all congenital birth defects. In the era of Sustainable Development Goals (SDGs) adopted by the United Nations in 2016, the focus on CHD should be integral to eliminating preventable child death and non-communicable diseases. These available global data were systematically analyzed providing both CHD mortality estimates and prevalence estimates. These findings were substantially based on a systematic literature review of different congenital anomalies to survey information on prevalence, associated mortality, and long-term health outcomes, in addition to subsequent estimates on disabilities. This study not only provides a more accurate assessment of the global burden of CHD, but also highlights the disproportionate disease burden in low Socio-Demographic Index (SDI) regions as compared to higher SDI regions.[6]

Additionally, as defined by the World Bank income classification, a significant number of children born with CHD worldwide have not received the medical or surgical treatment they need to survive in full health, since most live in low-and middle- income countries (LMICs). In 2020, the study by Vervoort et al. pointed out that nearly 26% of the population in LMICs is younger than 15 years of age, thus suggesting that as many 1.4 billion of children lack accesses to cardiac care if needed. In this study, the authors described certain progress in the field of global pediatric cardiac surgery with focus on 8 key stakeholders among advocacy efforts underlying these developments. Moreover, a framework was proposed to optimize civil society messaging to effectively advocate domestically and internationally to hold national and international policymakers accountable for existing gaps and disparities in CHD care around the world, in other words, a framework to increase interdisciplinary and cross-organizational collaboration.[7]

More recently, Tan and Aboulhosn, in their 2020 study on cardiovascular burden of coronavirus disease with a focus on CHD, have also indicated limited data on the effects of this infection on patients with cardiovascular disease, particularly so for those with CHD. Due to this infectious disease, extensive public-health measures to reduce person-to-person transmission of COVID-19 have been implemented globally to curb the spread of disease, reduce the burden on healthcare systems, and protect vulnerable populations. Their study summarized the early experience with COVID-19 and, in the future directions/next steps, also considered the potential applicability to and implications for patients with cardiovascular disease in general and CHD, in particular.[8]

Finally, the burden of childhood heart disease and inequality in access to pediatric cardiac care were brought to light by the publication of the four-part series of reports titled "The Invisible Child" by Children's Heart Link, in 2016. Furthermore, the concluding paper, "Voice for the Invisible Child: Childhood Heart Disease and the Global Health Agenda" claims that the leaders in global health development and local policymakers are partners in the increase of investments for sustainable and equitable access to pediatric care around the world.[9]

Taking into account all global efforts by a number of societies and nongovernmental organizations to accomplish the SDG) in CHD proposed by the United Nations in 2016, it is essential to accurately and systematically update these data on CHD burden in order to improve tracking of progress and encourage sustainable solutions to provide cardiac diagnosis and high-quality treatment for those with CHD. Strengthening the concept of burden in cardiac heart diseases, this study has chronologically shown, through a bibliometric analysis, the comprehensiveness of the term, pointing out major global inequities in CHD. It has also highlighted burden as a contributor to achieve proper care in CHD.

Methods

This study was outlined by the bibliometric technique. Bibliometrics is a statistical and quantitative method with the objective of presenting indices of production

and dissemination of scientific knowledge. It uses quantitative-citation analysis of publication data such as article-, author-, and journal-level data to determine and to demonstrate research productivity, quality, and impact.[10] More specifically, it includes analyzing the number of articles published by an author, the number of citations from those articles, and the ranking of the journals in which those articles were published. The use of bibliometric techniques is a common practice in research of sciences, since it enables qualitative and quantitative evaluation of scientific literature, allowing assessment of productivity within specific areas.[11] Its application helps in understanding of new issues and identifying trends for future research; that is, this method provides reflection on what has already been published, seeking theoretical propositions from the integration of theories that support the development of each theme, pointing out further research on the subject.[12]

Steps to carry out bibliometric analysis

The following steps can be used to design bibliometric analysis: definition of keywords; search for the terms in a chosen database source; insertion of search filters; data exportation; elaboration of a matrix; and exploratory factor analysis.[13]

Web of Science (WoS) database

The WoS Core Collection database source was chosen to carry out this bibliometric study, since it is one of the most widely used as a source for bibliometric research. It was originally produced by the Institute for Scientific Information, and it is currently maintained by Clarivate Analytics PLC (United States). Through WoS, tools are available for citation, reference, and h-index analysis, allowing bibliometric analysis. It covers approximately 12,000 journals. The subscription to this content offers the possibility to consult the following 5 collections: Science Citation Index Expanded (SCI-EXPANDED), with access availability from 1945 to the present; Social Sciences Citation Index (SSCI), with access availability from 1956 to the present; Arts & Humanities Citation Index (A&HCI), with access availability from 1975 to the present; Conference Proceedings Citation Index - Science (CPCI-S), with access availability from 1991 to the present; and Conference Proceedings Citation Index - Social Science & Humanities (CPCI-SSH), with access availability from 1991 to the present. It provides comprehensive citation data for many different academic disciplines and information about the number of citations of each article, journal relevance, among others. The Core Collection includes 79 million records, and the Platform includes 171 million, with temporal coverage from 1900 to present. It covers disciplines such as science, social science, arts, and humanities, supporting 256 disciplines. WoS tends to favor English-language publications from the United States that are peer reviewed and have a long publication record.[14]

Operationalization

Afterwards, the keywords summarized as a title were defined to carry out the bibliometric analysis and inserted into the database chosen for the search as follows: "Burden CHDs." This was important for addressing the main theme of the study. Initially, a total of 65 publications were found using these keywords as a title (keywords summary). Subsequently, it was necessary to define the search filters produced and presented by the WoS database, such as: publication years, WoS Categories, document types, consolidated organizations, research areas, authors, countries and regions, English Language, and whether the search will include only articles published in periodicals or in annals of events. Chronologically, bibliographic references and related authors were also surveyed. After surveying the years of publication (2005 to 2022), the last decade was chosen to filter some relevant articles, since the highest number of publications on this issue was released during these years (2010 to 2022). All these filtered searches were grouped and presented in figures showing the main results that were considered important in this period.[15]

Finally, submission of this study to the Ethics Committee on Human Research (CEP/FAMERP) was not required, according to article VI, Resolution number 510/16 (2016) of the Brazilian Ministry of Health, National Health Council, which states that evaluation or registration by the CEP/CONEP system is not required for research that uses information in the public domain, such as this study, which was performed exclusively with scientific texts for scientific literature review. The access to the chosen database of medical literature to be surveyed was available through the institutional subscription, access by: Faculdade de Medicina de São José do Rio Preto (FAMERP), São Paulo, Brazil; Portal de Periódicos Capes (CAPES MEC); acesso remoto via Comunidade Acadêmica Federada, CAFe; Buscar Base: WoS - Coleção Principal (Clarivate Analytics) Therefore,

free access to survey all required publications was made available to the researchers.[16,17]

Results

Results are presented as figures according to each topic investigated, namely, year of publication, countries and regions, rank by citation, and document types, as well as a central Figure.

Figure 1 shows 65 publications between 2005 and 2022, with greater emphasis on the year 2022.

Figure 2 displays 28 countries where studies were carried out and published.

Figure 3 shows the ranking of authors and the number of citations with 200 authors and 270 citations, due to 49 authors having more than 1 citation.

Figure 4 presents the citation analysis and shows the 10 most highly-cited articles and their references, which is a useful way to assess the impact or influence of these articles in the field of CHD.

Figure 5 describes the division of the most represented fields of publication in the WoS.

Figure 6 displays the types of documents published, such as articles, meeting abstracts, letters, review articles, corrections, editorial materials, and conference communications.

Figure 7 cites the research areas in which the concept of burden was found, Cardiovascular System and Cardiology being the most frequent.

Discussion

The repercussion of the article Global, Regional, and National Burden of CHD, 1990-2017: A Systematic Analysis for the GBD Study 2017,[3] was considered a milestone in the field of health care worldwide.

This present study has performed a survey about the concept of burden in CHD considering it as a contributor to achieve SDGs. The article Global, Regional, and National Burden of CHD, 1990-2017: a Systematic Analysis for the GBD Study 2017 indicated the concept of burden in CHD globally, regionally, and nationally. The article was published in March 2020, which was the year with highest number of publications, according to the results presented in this study (Figure 1, Table 2).

More recently, the article Global, Regional, and National Time Trends in Mortality for Congenital Heart Disease, 1990-2019: an Age-Period-Cohort Analysis for the Global Burden of Disease 2019 Study by Su et al. was published in January 2022 (Figure 1), with 50 references (Figure 4). This article reinforces the relevance of this subject being approached up to these recent times.

This bibliometric analysis performed using the WoS database has proposed to investigate since when the concept of burden has been related to CHD. According to the results, the main selected points for discussion are year of publication, countries and regions, authors, most cited works, and disagreement between authors.

Years of publications

At first, taking into account the main objective of this study, that is, to chronologically investigate the concept of burden in CHD, years of publications were refined, totaling 14 years (Figure 1). The first 2 publications were found in the year of 2005; since then, no publications were found in the years of 2006, 2008, and 2014. In the last decade, from 2011 to 2021, the amount was expressive, mainly in the year of 2020 with 11 (the highest number); 6 in the year of 2021, and 1 in early 2022 (Figure 1).

Countries and Regions

Publications were from 29 countries and regions. The United States of America, Canada, England, and Netherlands were the countries with the highest number of publications. This indicates that these countries can be considered excellent in the care of CHD. The People's Republic of China and India are regions with expressive numbers of publications. Through Jae Young Lee's 2020 publication, Global Burden of CHD in Korea as a Potential Solution to the Problem[8] stands out, since CHD is not currently a "burden" in the country. Other countries presented 2 articles; Brazil has 1 publication, although the authors of the publication were from China (Figure 2).

Authors and most cited articles

Two thousand authors comprised the authorship of the 65 publications, counting from the first author to the other contributors (*et al.*). It is important to note the most cited authors during the period from 2005 to

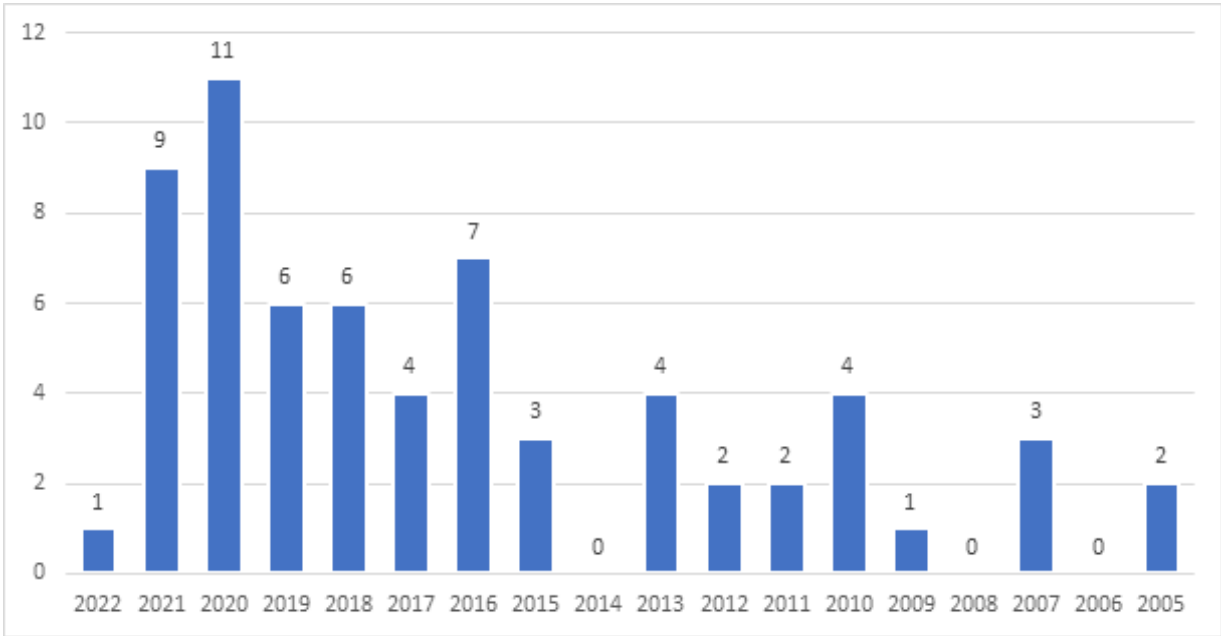

Figure 1 – Year of publication: Chronologically, publications from 2005 to 2022, with the highest number in 2020.

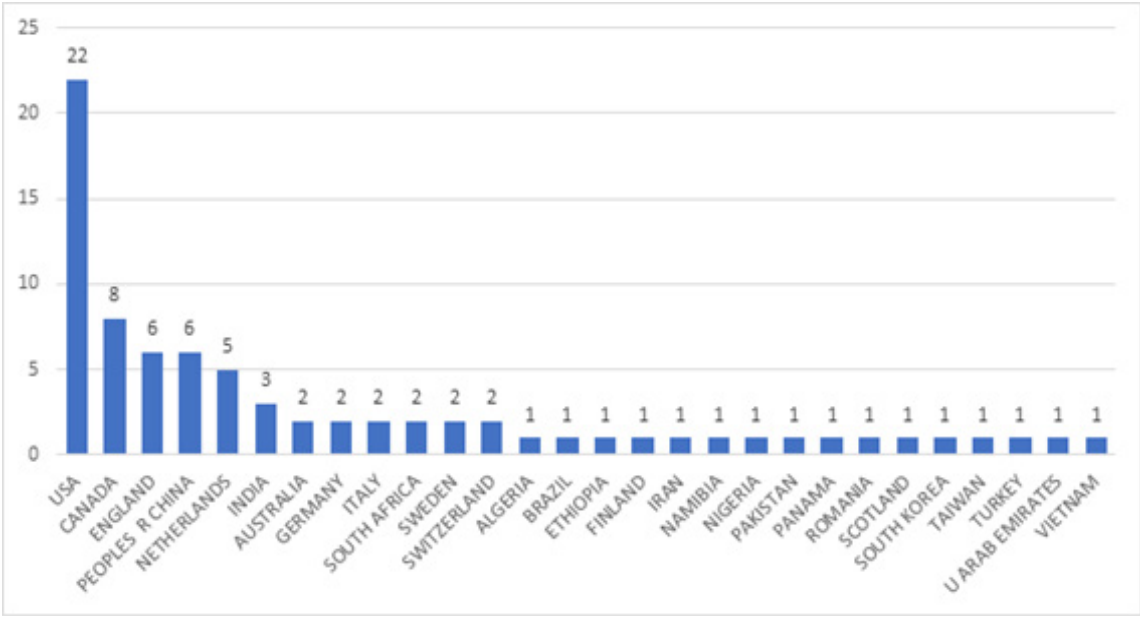

Figure 2 – Countries and regions: The USA produced the highest number of publications by country/region. Brazil had 1 publication.

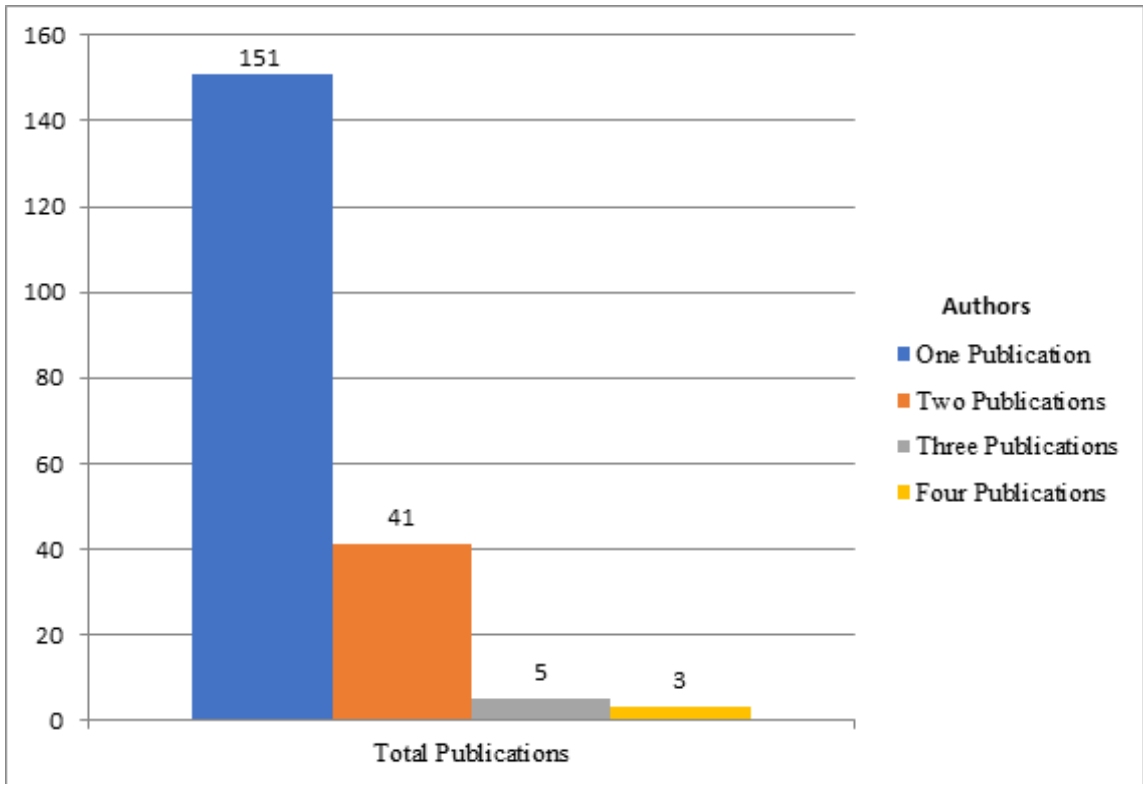

Figure 3 – Authors and number of publications.

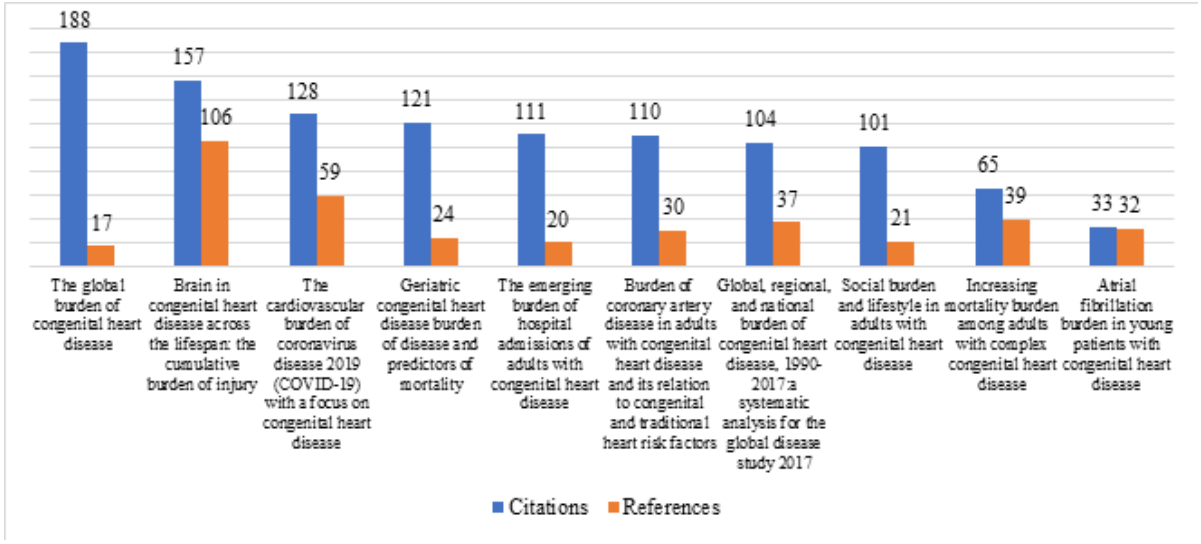

Figure 4 – Ranking by number of citations: The 10 most highly-cited articles and their references, a useful way to assess the impact or the influence these articles had in the field of CHDs.

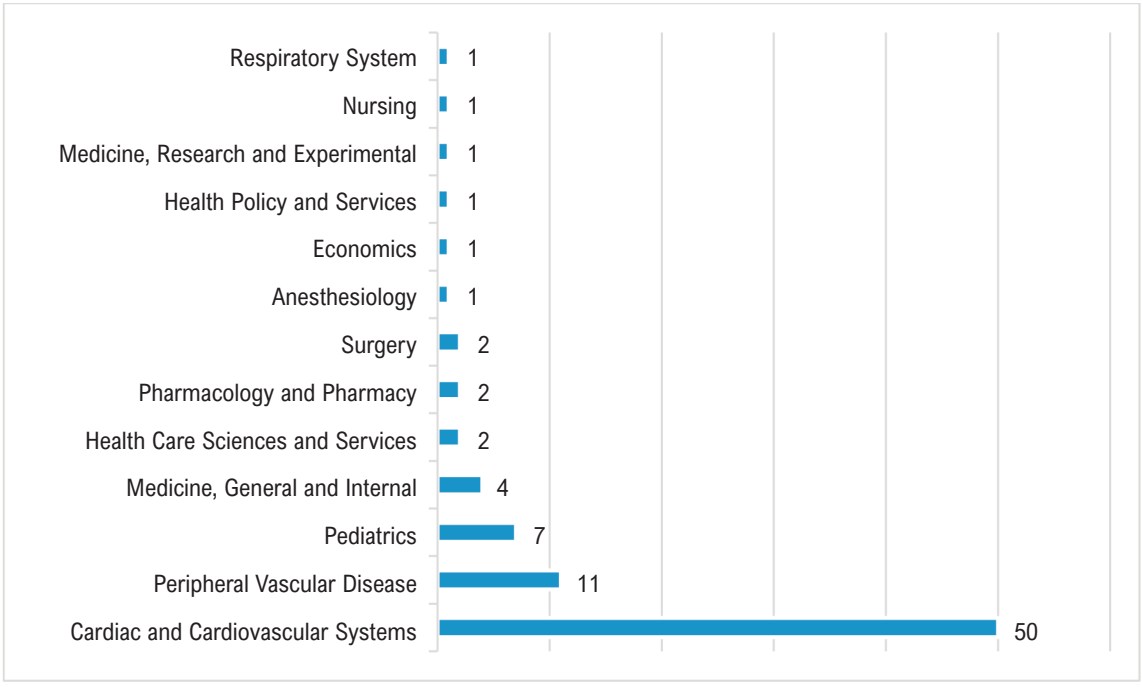

Figure 5 – WoS categories: Division of the most represented publication fields.

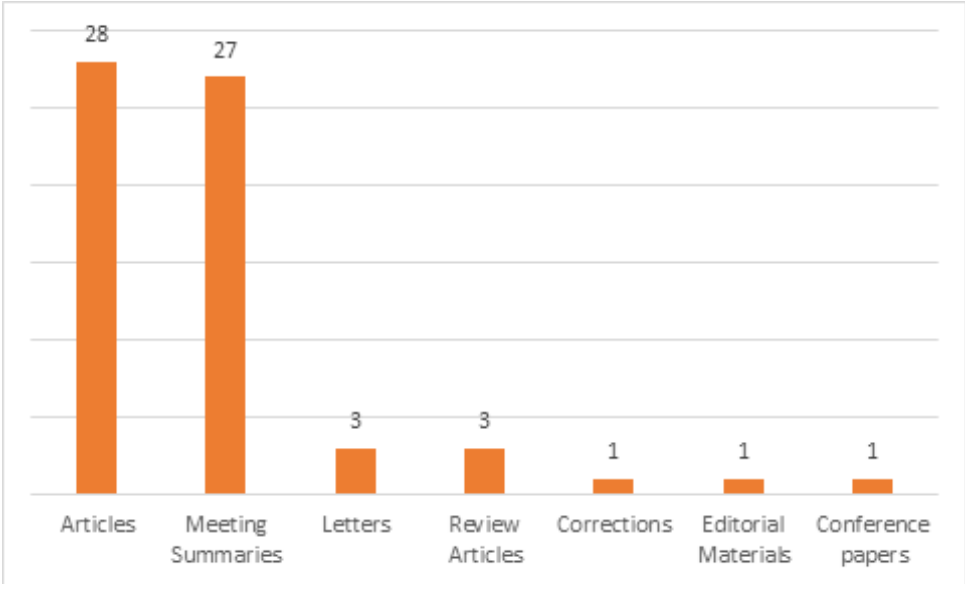

Figure 6 – Document types

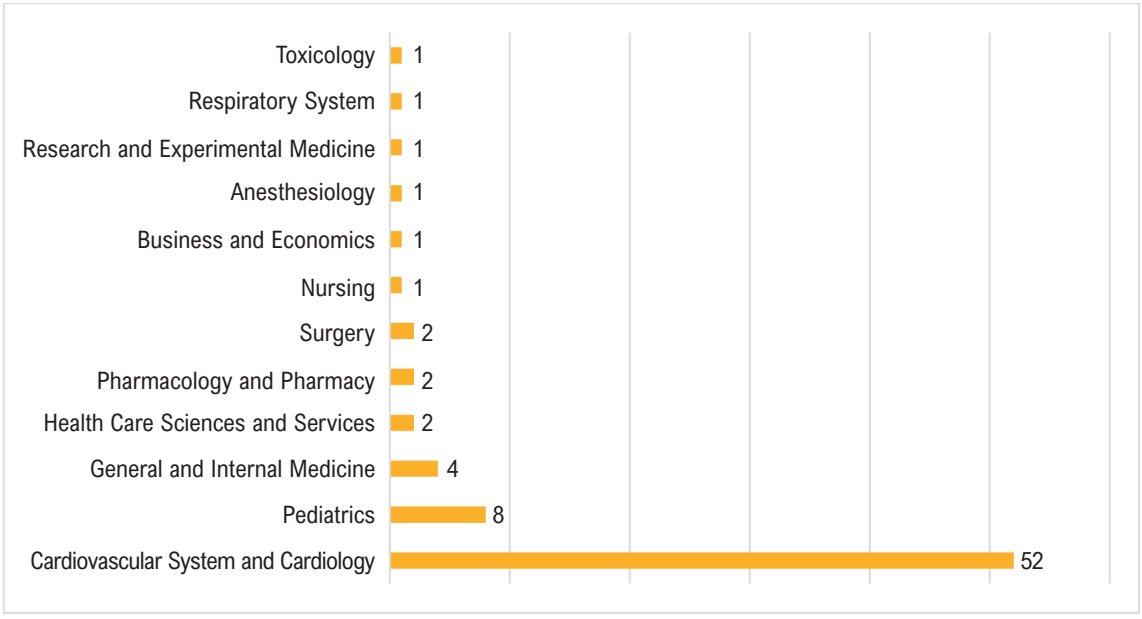

Figure 7 – Research Areas: The concept of burden was most frequently found in the area of Cardiovascular System and Cardiology.

2022, namely, Hoffman, Tan, and Aboulhosn, from the United States. "The global burden of CHD," published in the *Cardiovascular Journal of Africa* had 188 citations and 17 references. The position of these first authors was 57%. "Brain in CHD Across the Lifespan: The Cumulative Burden of Injury" by Marelli et al. was the second in the ranking by citation, with 157 citations and 106 references. "The cardiovascular burden of coronavirus disease 2019 (COVID-19) with a focus on CHD" by Tan and Aboulhosn was third, with 128 citations and 59 references (Figure 4).

No tables or figures were elaborated comprising the 65 articles since 10 of them were considered according to the highest number of publications as well as countries and most cited.

Disagreement between authors

It is worth highlighting Tan and Aboulhosn's discussion[8] with Müller et al.[10] regarding their disagreement that burden of CHD was not only in times of COVID-19. A letter to the editor by Müller et al. reported this disagreement, with an immediate response from Tan and Aboulhosn, "Response to the letter: The cardiovascular burden of CHD - not only in times of Covid-19".[10]

Limitations of the study

As this analysis was carried out through a single database (WoS), no comparison was established with other databases, resulting in a certain limitation of the study.

Conclusion

This bibliometric study performed using the WoS database has indicated that all 65 publications have strengthened the concept of burden addressing a concern in CHD. This concept has been used with great propriety in relation to achieving available goals regarding the care of CHD, mainly in the last decade, with the year of 2020 standing out.

Author Contributions

Conception and design of the research, analysis and interpretation of the data, statistical analysis and critical revision of the manuscript for intellectual content: Sciarra AMP, Cobo DL, Batigalia F, Croti UA; acquisition of data: Sciarra AMP; writing of the manuscript: Sciarra AMP, Cobo DL, Batigalia F.

Potential Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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Study Association

This study is not associated with any thesis or dissertation work.

Ethics Approval and Consent to Participate

This article does not contain any studies with human participants or animals performed by any of the authors.

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