



## Cost-effectiveness of Breast Cancer Screening from 2013-2022: A Bibliometric Study

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### ABSTRACT

**Background:** Breast cancer in women is the most common cancer case worldwide. Research on the cost-effectiveness of breast cancer screening still needs to be studied in depth and broadly. This study aims to show the development of research on the most effective and efficient breast cancer screening (2013-2022). **Methods:** This is a qualitative literature study that used Scopus database. **Keywords:** (Breast Cancer) AND (Screening) AND (Cost Effectiveness). 459 articles can be obtained from a total of 2.108. Subject area is related to medical scope, the document type is an article and journal in English language. This research uses VOS viewer and R-Studio for analysis. **Result:** The number of publications on this topic are fluctuating, the most publications in 2021 (61 articles). The 3 most authors are De Koning HJ, Van Reverstyn NT and Algoz O, affiliated with University of Toronto, University of Manchester and Harvard Medical School. Value In Health journals contributed the most publications. The most keywords: Female by 622 times. After 2021 the most discussed themes relate to human tissue, willingness to pay, and clinical outcomes. **Conclusion:** It is necessary to involve many LMICs in this topic research to obtain more applicable screening methods and modalities in countries with limited health facilities. The scope of research that is still open includes CEA research in LMIC, breast cancer in men, the quality of life of breast cancer patients, patient outcomes, genetic screening-testing and also gene expression profiling.

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### INTRODUCTION

Breast cancer in women is the most common cancer case worldwide. The Global Cancer Observatory estimates that the number of new cases in 2020 will be 2.3 million cases (Sung.H., 2021; Curado., 2011). Breast cancer is also the most common cause of death in women with cancer, which is 1 in 6 women who suffer from cancer. The incidence rate in high-income countries (HICs) is 88% higher than that in low-to-middle-income countries (LMICs) (55.9-29.7 age-standardized rates per 100 000 women, respectively) (Momenimomaved., 2019). However, it turns out that the death rate in countries with low-middle income per capita is reported to be 17% higher than in HICs countries (15.0-12.8 age-standardized rates per 100,000 women, respectively). one is because countries with HICs have better healthcare infrastructure (Curado., 2011; Harton S.,2015).

The World Health Organization's (WHO) Global Action Plan for the Prevention and Control of Noncommunicable Diseases (NCDs) 2013 to 2020 listed 6 objectives and 9 voluntary global targets for the interventions of NCDs (WHO., 2020). Countries involved in the plan were to work on creating effective programs to reduce and eventually eliminate the number of NCD cases, including cancer. The WHO Independent High-level Commission on Non-communicable Diseases delivered a list of 8 further recommendations to the WHO to assist in the curbing of NCDs (WHO., 2020). The involved nations were encouraged to engage with their respective societies to prevent the spread of NCDs, which include cancer, by ensuring that their people received the necessary information to make healthy choices.

The morbidity and mortality rates of cancer in LMICs can be improved by implementing various cancer awareness programs, prevention strategies, and early diagnoses and

treatments (Curado., 2011; Ebell., 2018; Shah SC.,2019). Issues with cancer control in LMICs include the (1) uneven allocation of health resources, (2) uneven access to healthcare, and (3) low national budgetary allocation to health services (Curado., 2011; Rivera., 2018; Horton.,2015).

The survival rate of breast cancer for at least 5 years after diagnosis ranges from 90% in HICs to 66% in India and 49% in Malaysia (Curado., 2011; Abdulah.,2011; Norsa' dah.,2011). Early detection and treatment have been proven successful in decreasing breast cancer mortality rates in HICs and should be applied in LMICs where some of the standard tools for detection and treatment are available. The late screening and diagnosis of breast cancer among women are prevalent in LMICs compared with developed countries (Rivera.,2018; Horton.,2015).

The resources allocated to public health are generally limited and must be used effectively to achieve optimal outcomes. An analysis of the cost effectiveness of breast cancer prevention and screening programs is essential to demonstrate their value, particularly in LMICs. Health economics play an important role in the decisions made regarding resource allocation by providing evidence-based cost-effectiveness analyses (CEAs) of cancer prevention programs (WHO.,2022).

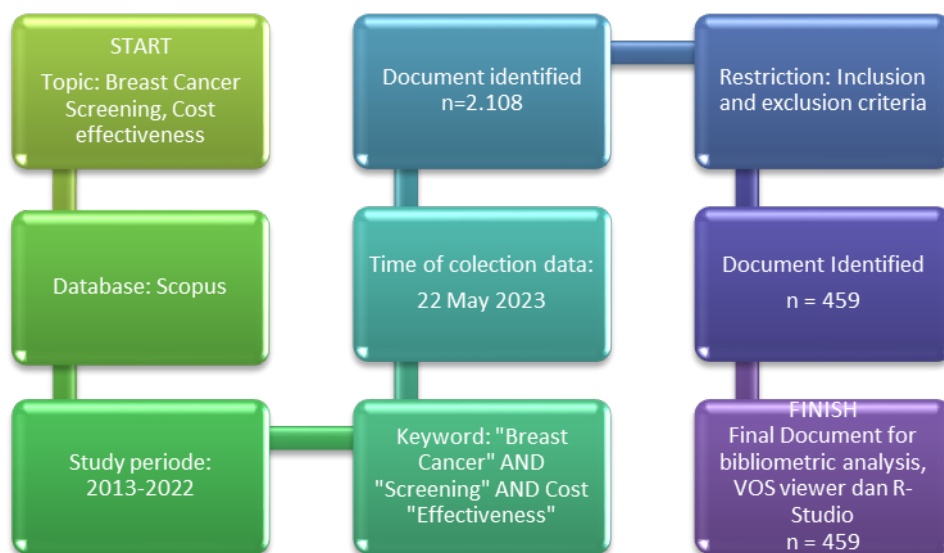
Research on effective and efficient breast cancer screening modalities is still very limited and depends on a country's economic conditions which greatly influence the health facilities it has. Research on the cost-effectiveness of breast cancer screening still needs to be studied in depth and broadly to provide recommendations for early detection of breast cancer that can be used effectively and efficiently in various countries, especially poor and middle-income countries. This study uses bibliometric methods to describe qualitatively and quantitatively research trends and

scientific literature, authors, impact of publications, citation analysis, analytical journals, countries that contribute to the research field.

This study aims to show the development of research on the most effective and efficient breast cancer screening modalities from 2013-2022. In this study the authors used bibliometric analysis and VOS viewer version 1.16.18 and R studio. Documents and articles analyzed were obtained from Scopus. This analysis is important because it provides insight into the scope of existing research and the potential for it's development.

## METHODS

This research method uses Scopus as an article source search database. As we all know, Scopus is a reputable and trusted journal database provider, especially in the field of health and therapy. The method used in the search uses the following keywords: *(Breast Cancer) AND (Screening) AND (Cost Effectiveness)*. Based on the strategy of using these keywords, in the early stages, 2,108 articles were found from 2013 to 2022. Then after applying the inclusion and exclusion criteria, data was obtained for 459 articles. The inclusion criteria for this panel were all journals in the form of articles and reviews; final publication; English language journal and subject area limited to medical scope. While the exclusion criteria are articles that use keywords that are not related to breast cancer (*economics, colorectal cancer, ovary cancer, uterine cervix cancer, Prostate cancer, lung cancer, ovarian neoplasm, ovary tumor, salpingoophorectomy, colon cancer*). The data collection strategy is illustrated in the flowchart in **Figure 1**



**Figure 1. Data Search Pitfall related to Breast Cancer Screening Cost effectiveness.**

Some information was collected from articles that met the inclusion and exclusion criteria set by the authors. This information includes published data every year, year of publication, keywords, author and affiliation data and countries that contribute to the research field. Bibliometric analysis was performed using *VOS viewer and R-Studio*

*Biblioshiny. VOS viewer and R-Studio Biblioshiny* used to provide a better visualization of the data obtained; whether in the form of graphs, diagrams or network between article data so that it is easier to interpret (Pan X.,2018; Guleria., 2021).

## RESULT AND DISCUSSION

Based on the database obtained from Scopus, basic data is obtained from 459 articles which will be analyzed using the Vos viewer. The analysis includes the year of publication of the article, author data, countries that contributed to the research field, contributing institutions and article document affiliations.

### Result

As for basic data, general information about the articles analyzed in this study is presented in table 1. The trend in number of publications regarding the cost-effectiveness breast cancer screening each year from 2013 to 2022 can be seen in Figure 2. The graph shows that the number of publications on the topic of cost-effectiveness breast cancer screening research are still very fluctuating. The most publications seen in 2021 were 61 published articles. In 2015, the number of publications was quite high, namely 53 articles compared to 2013 (49 articles) and 2014 (38 articles). After 2015, the number of publications continued to decline until it was lowest in 2018 (34 articles) and then began to rise again in 2019 until it reached its highest rate in 2021. Meanwhile, in 2022 the number of publications decreased again, with 53 articles published.

Another results of the bibliometric analysis are related to the distribution of authors in publishing articles about the cost-effectiveness breast cancer screening. Data were analyzed based on the number of publications and the year of publication. In Figures 3a and 3b it shows that the 3 most authors on the research topic are De Koning HJ with 16 articles, Van Reverstyn NT (11 articles) and Algoz O (10 articles). Wang J, De Bock GH and Greuter M.J.W have a relatively smaller number of publications compared to De Koning HJ, Van Reverstyn.NT and Algoz.O but have a relatively newer publication.

Based on published data about affiliated institutions on the topic of research on the cost-effectiveness breast cancer screening, it was noted that most of the authors were affiliated or came from University of Toronto (25 articles), followed by University of Manchester (24 articles)) and Harvard Medical School (22 articles). Figure 4b also shows that the 3 institutions are very consistent as affiliated institutions for authoring articles on cost-effectiveness of breast cancer screening, where the number of publications continues to show an upward trend from year to year.

Figure 5 presents the author distribution data based on the author's country. Most of the authors come from the USA, the Netherlands, the United Kingdom, China and Canada. Total number of countries contributing to publications regarding the cost-effectiveness breast cancer screening is 52 countries. Table 2 shows that in the 2013-2022 period, the majority of authors came from the USA with 131 articles and MCP ratio of 0.19. The Netherlands with 32 articles with MCP ratio 0.406 is in second place. In third place is United Kingdom with 32 articles and an MCP ratio of 0.218; followed by China and Canada with 22 articles and 21 articles respectively with MCP ratios of 0.318 and 0.238.

The results of the analysis from the journals that published the most articles on topics according to this study can be seen in Figure 6. Value In Health journals published 13 articles that placed them in first position. In second place is BMC CANCER with 11 published articles, followed by the American Journal of Roentgenology with 8 articles. The International Journal of Cancer has published 8 articles. Breast Cancer Research and Treatment with 7 articles.

Meanwhile, the Annals of Internal Medicine, the Asian Pacific Journal of Cancer Prevention published 6 articles by each of them.

Based on the data that occurs in Figure 7, it can be interpreted that there are 10 journals that are most widely cited on the topic of research on the cost-effectiveness breast cancer screening between 2013-2022. Cochrane Database Sys Rev is the journal that most cited by many researchers with a total 833 times in articles written by Stacey.D (2014); 740 times cited in article written by Stacey D. (2017). Next in line is New England J Med with 575 citations, followed by J.AM Chem SOC with 465 citations.

Analysis of the most used keywords in searching articles according to research topics using Biblioshiny can be seen in Figure 8 and Table 3. The results of this analysis show that the most used keywords include: Female by 622 times, human 462 times, mammography 413 times, breast neoplasm 369 times, humans 352 times, breast cancer 331 times, and cost effectiveness analysis 292 times.

Figure 9 and Figure 10 depicts a network visualization of research on cost-effectiveness breast cancer screening, in which 550 related word items were obtained with a link count of 53,098 and a total link strength of 196,390. The themes that have been widely researched before 2018 are related to human, female, breast cancer, mammography, middle age, breast neoplasms, and early diagnosis. While research around 2020 uses several topics such as breast density, digital breast tomosynthesis, breast tumors etc. Whereas after 2021 the most discussed themes relate to human tissue, willingness to pay, and clinical outcomes.

Figure 11 shows the density level indicated by the number of frequently appearing keywords in bright yellow; as seen on the label human, breast cancer, cost effectiveness, breast tumor and others. Then, the bright green color that surrounds it labels cancer risk, breast density, quality adjusted life year, diagnostic imaging and others. These areas are topics that have been extensively researched. While the topics covered in dark green and blue are those that are still a little bit, such as genetic testing, genetic screening, BRC protein and others.

**Table 1. Main Information related research data**

| MAIN INFORMATION ABOUT DATA     | RESULT    |
|---------------------------------|-----------|
| Timespan                        | 2013:2022 |
| Documents                       | 459       |
| Annual Growth Rate %            | 0.88      |
| Document Average Age            | 5.35      |
| Average citations per doc       | 26.43     |
| References                      | 23165     |
| <b>DOCUMENT CONTENTS</b>        |           |
| Keywords Plus (ID)              | 3730      |
| Author's Keywords (DE)          | 978       |
| <b>AUTHORS</b>                  |           |
| Authors                         | 2748      |
| Authors of single-authored docs | 25        |
| <b>AUTHORS COLLABORATION</b>    |           |
| Single-authored docs            | 29        |
| Co-Authors per Doc              | 7.06      |
| International co-authorships %  | 27.23     |
| <b>DOCUMENT TYPES</b>           |           |
| article                         | 334       |
| review                          | 125       |

Source: R-Studio Biblioshiny

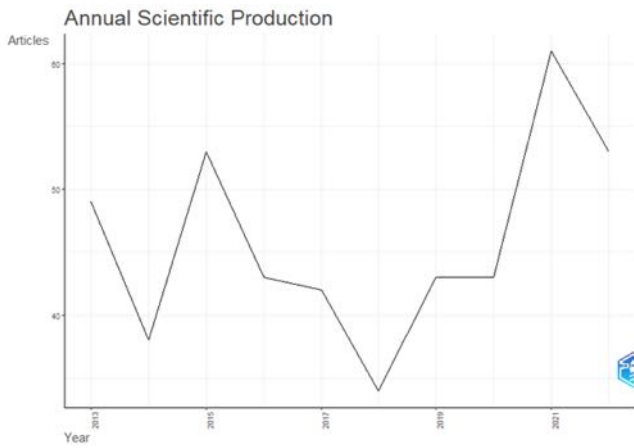


Figure 2. Publication trends of the cost effectiveness on Breast cancer Screening. Source: R-Studio Biblioshiny

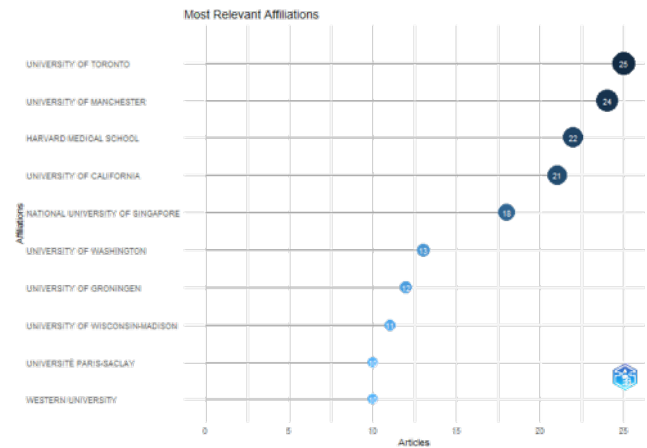


Figure 4a. Publication based on the author's institution of the theme cost effectiveness breast cancer screening. Source: R-Studio Biblioshiny

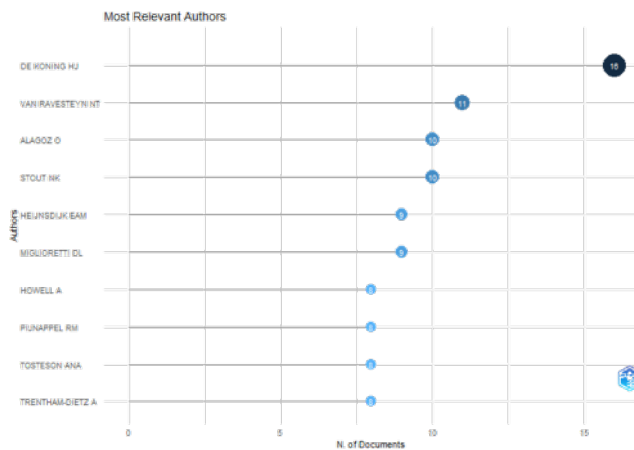


Figure 3a. Publication based on most author's. Source: R-Studio Biblioshiny

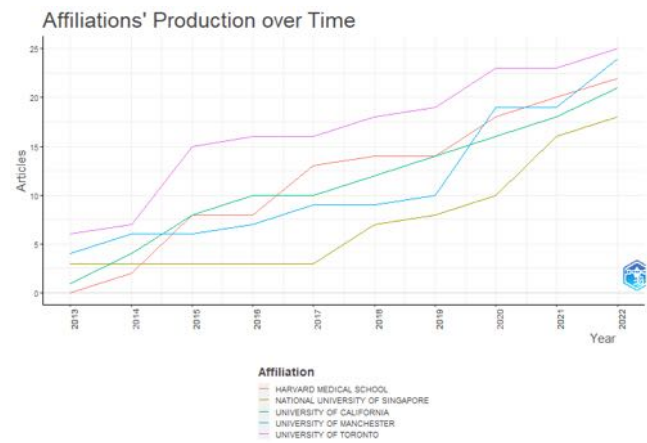


Figure 4b. Productivity trends of authors Affiliation from 2013-2022. Source: R-Studio Biblioshiny

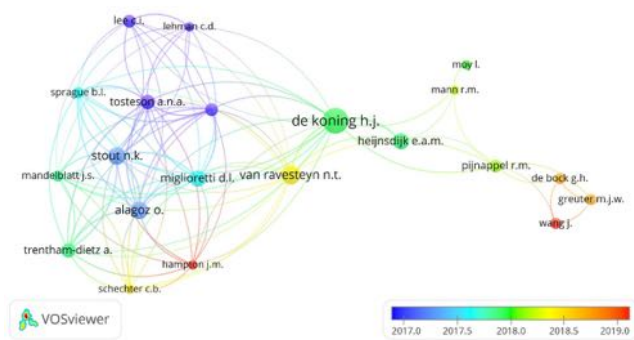


Figure 3b. Distribution of authors of articles on cost-effective early detection of breast cancer based on number and year of publication 2013-2022. Source: VOS Viewer

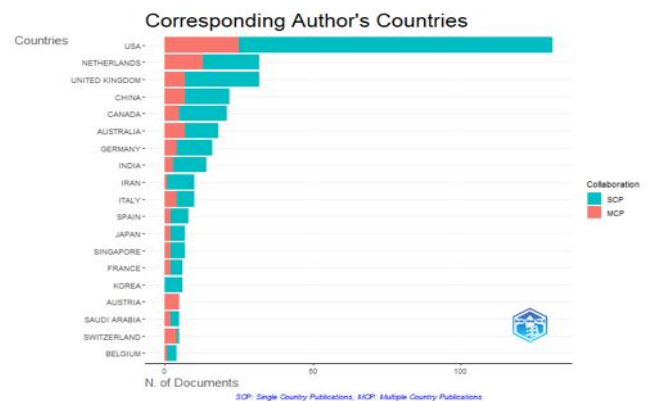


Figure 5. Correspondence by author country. Source: R-Studio Biblioshiny

Table 2. Cosrrspondence by country of author

| Country        | Articles | SCP | MCP | Freq  | MCP_Ratio |
|----------------|----------|-----|-----|-------|-----------|
| USA            | 131      | 106 | 25  | 0.285 | 0.190     |
| NETHERLANDS    | 32       | 19  | 13  | 0.069 | 0.406     |
| UNITED KINGDOM | 32       | 25  | 7   | 0.069 | 0.218     |
| CHINA          | 22       | 15  | 7   | 0.047 | 0.318     |
| CANADA         | 21       | 16  | 5   | 0.045 | 0.238     |
| AUSTRALIA      | 18       | 11  | 7   | 0.039 | 0.388     |
| GERMANY        | 16       | 12  | 4   | 0.034 | 0.25      |
| INDIA          | 14       | 11  | 3   | 0.030 | 0.214     |
| IRAN           | 10       | 9   | 1   | 0.021 | 0.1       |
| Italy          | 10       | 6   | 4   | 0.021 | 0.4       |

Source: R-Studio Biblioshiny

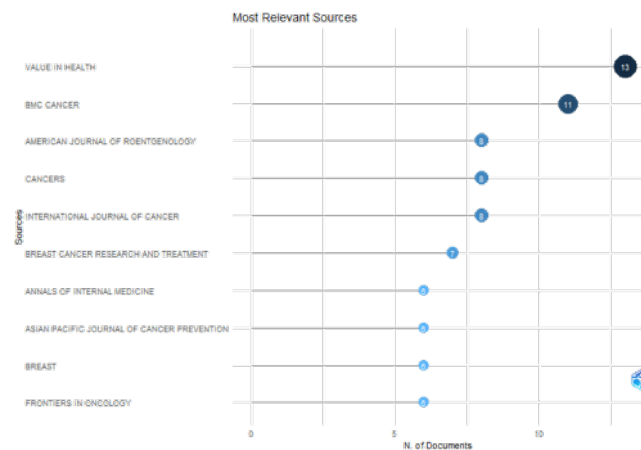


Figure 6. Journals that published the most articles on the cost effectiveness breast cancer screening. Source: R-Studio Biblioshiny

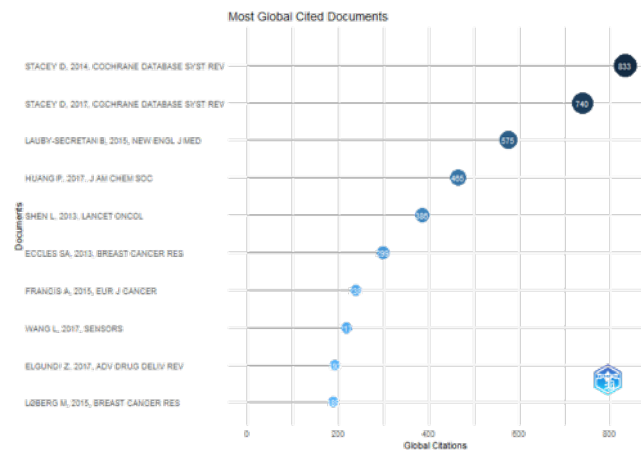


Figure 7. The most cited journals in research on cost effectiveness breast cancer screening. Source: R-Studio Biblioshiny



Figure 8. The most common words found in the research study of cost effectiveness breast cancer screening. Source: R-Studio Biblioshiny

Table 3. The most common keywords

| Terms                       | Frequency |
|-----------------------------|-----------|
| female                      | 622       |
| human                       | 462       |
| mammography                 | 413       |
| breast neoplasms            | 369       |
| humans                      | 352       |
| breast cancer               | 331       |
| cost effectiveness analysis | 292       |

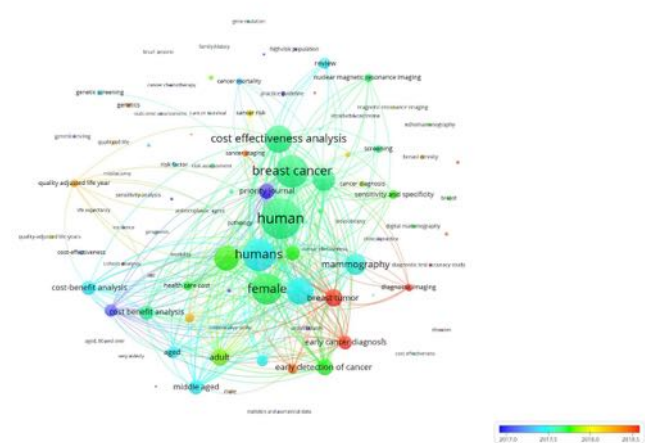


Figure 9. Overlay visualization of the Cost effectiveness breast cancer screening. Source: R-Studio Biblioshiny

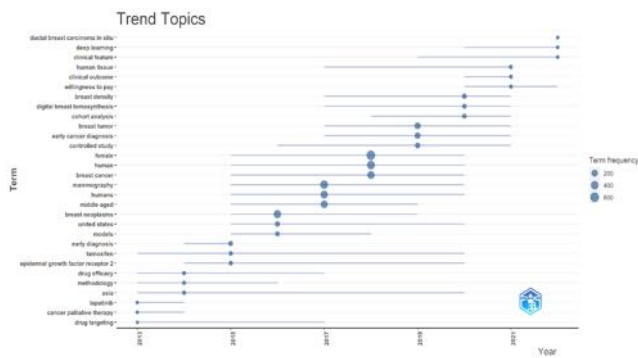


Figure 10. Trend topics related to cost effectiveness breast cancer screening research. Source: R-Studio Biblioshiny

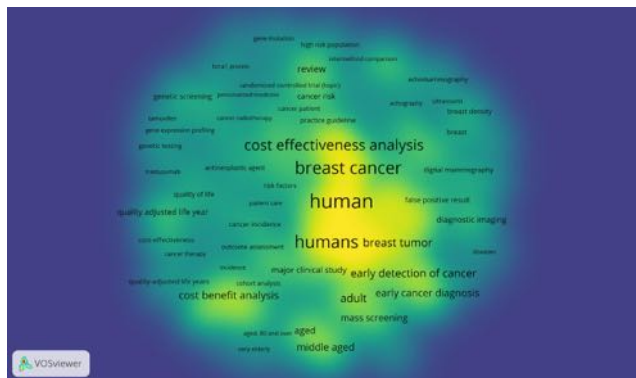


Figure 11. Density mapping related to cost effectiveness breast cancer screening. Source: VOS Viewer

## DISCUSSIONS

In accordance with the objectives of this study and based on the results of the bibliometric analysis above, several aspects can be discussed so that they can provide a more in-depth scope of research that has been conducted regarding cost-effectiveness of breast cancer screening. In general, from 2013–2022 the number of studies discussing this research topic has increased. Even so, the increase in scale is still low with a growth rate of 0.88% per year. In addition, the number of articles produced each year is still fluctuating, with an average of 45 articles per year; the highest number in 2021 (63 articles) and the lowest in 2018 (34 articles). This indicates that research in the field of cost effectiveness breast cancer screening still needs to be improved considering the methods and technology for breast cancer screening continue to develop.

In terms of authors, the majority of authors come from the USA, the Netherlands, England, China and Canada. These countries are developed countries with high economic levels, so the technology and health facilities they have are also better. The problem that occurs is that not all countries, especially in countries with low economic levels and developing countries, have adequate health facilities. Breast cancer screening methods that are considered effective and efficient in developed countries may not necessarily be applicable in developing and low-income countries (Al Foheidi.,2013). So, it is necessary to involve many of these countries in research on cost effectiveness of breast cancer screening in order to obtain screening methods and modalities that can be more applicable in countries with

limited health facilities. In line with this, when looking at the MCP ratio (*Multiple Country Publication*) which is still low in the 5 most productive countries. It is hoped that in the future there will be more collaboration between developed countries and underdeveloped and developing countries on this research topic. This can also encourage underdeveloped and developing countries to learn more not only about the development of breast cancer diagnostic technology, breast cancer therapy, quality of life for breast cancer patients, but also the health systems of developed countries.

Results of bibliometric analysis on published journal sources, *Value in Health* is the source of the most published journals on the topic of cost-effectiveness breast cancer screening, followed by *BMC cancer dan American General of Roentgenology*. On the journal website known that *Value in Health* is an indexed international journal which publishes original research and articles that primarily concentrate on health policy advancing the field of health economics and research outcomes to help healthcare leaders make evidence-based decisions. *Value in Health* is a monthly publication distributed to 10,000 readers worldwide. Over the last few years, *Value in Health* has shown significant growth in journal impact factors. The journal's current impact factor is 5,725 and its 5-year impact factor score is 6,932. The journal is now ranked 4th out of 88 journals in health sciences and services, 9th out of 108 journals in health policy and services, and 24th out of 376 journals in economics (social sciences) (Value In Health.,2023).

Another aspect to be analyzed is the author's relationship. The three authors who published the most articles on cost effectiveness breast cancer screening were De Koning HJ, Van Revesteyn NT, and Algoz O out of a total of 2,748 authors who contributed to the research topic from 2013–2022. The majority of the 3 authors mostly wrote about methods and modalities of breast cancer screening with settings in developed countries such as the Netherlands, America, England, Norway and so on. There is still little research on the most cost effectiveness screening methods and modalities to be implemented in low-to-middle-income countries (LMICs). The adoption of various screening/early detection programs for breast cancer is motivated by the desire to reduce mortality rates and has the highest cost-effective value (Bertram., 2017; Jayasakera., 2020). The modality of mammography examination is the most widely discussed modality by these researchers. This modality is considered the most cost-effective for breast cancer screening in developed countries. However, low-to-middle-income countries (LMICs) cannot adopt easily without very careful consideration (Rashidian.,2013; Luckman., 2019; Yoo KB.,2013). This is due to the limited resources in the health sector and there is still a need for cost-effective studies to provide guidance to them in planning and implementing breast cancer detection and screening programs in each country (Rashidian.,2013; Luckman., 2019; Lee M.,2017). In the last 10 years, studies on *Effective cost analysis (CEA)* on the topic of early detection of breast cancer in the *LMICs* recorded is still very small compared to developed countries. CEA studies conducted in LMIC countries show different results with influencing factors depending on the screening method used, the incidence rate of breast cancer, and the conditions at the geo-socio-economic level in each country (Sun L.,2017; 2019). From this, the scope of research that is still open to be carried out is research on CEA in LMICs countries and studies that compare differences in the allocation of health resources in each LMICs to mortality and morbidity rates from breast cancer.

This bibliometric analysis also discusses the keywords that appear the most on the research topic of cost-effective breast cancer screening from 2013-2022. From this analysis, one can see the direction of development and trends in the topics that have been studied. The results of the bibliometric analysis of the words that appear a lot on the research topic include: *female*, *human* and *mammography*. *female* and *human* are the word that appears the most because it is known that the majority of sufferers are women. Although the incidence rate is small, breast cancer can also occur in men (<1% of breast cancers, <1% of cancers in men and 1 patient in 100,000 men). Men with a family history of breast cancer are 2.5 times more likely than men who do not have a family history of breast cancer. Relatively few people know about breast cancer in men, such as etiology, signs and symptoms and how to detect cancer early, due to the rarity of this disease so that more than 80% of cases are found at an advanced stage (Komite penanggulangan Kanker Nasional., 2013; Silvestri., 2016; Mathew., 2008). The topic of research on breast cancer in men also still opens up opportunities for research.

*Mammography* is also a word that is often mentioned in research journals on the topic of cost-effective breast cancer screening because it is one of the most widely discussed screening modalities. However, there are still many examination modalities that need to be further investigated regarding their effectiveness, such as MRI, tomosynthesis, USG, FNAB, biomarkers and combinations of each of these examination modalities. In addition, the approach method used also influences the effective cost of a screening program, whether the approach is for at-risk populations with specific age and genetic factors or based on the entire population in an area (Bimbaum., 2018).

Density on topics that are widely discussed in research can be presented in the form of density exposure images from the results of analysis using *VOS Viewer*. In this description, it can be seen that the research topics in areas with yellow notifications are topics that have been widely discussed. While topics that are in areas with green notifications are topics that are still rarely researched. From the results of this analysis there are many topics that still need to be developed and become opportunities for research, for example those related to the quality of life of breast cancer patients, patient outcomes, genetic screening, genetic testing and also including gene expression profiling.

## LIMITATION OF THE STUDY

The limitation of this study is that the source of the articles/journals used is only from one source, namely Scopus, so the number of journals obtained from the keywords set by the authors is relatively small, so the information data collected is less comprehensive.

## CONCLUSION AND SUGGESTION

In general, from 2013-2022 the number of studies discussing the topic of cost effectiveness research on breast cancer screening has increased, although it is still fluctuating and the growth rate is still low (0.88% per year). De Koning HJ, Van Revesteyn NT, and Algoz O are the 3 most prolific writers; the majority of authors come from developed countries with research settings also in developed countries, so it is necessary to involve many LMICs in cost effectiveness

breast cancer screening research in order to obtain screening methods and modalities that can be more applicable in countries with limited health facilities. The scope of research that is still open includes CEA research topics in LMIC countries, differences in the allocation of health resources in each LMIC country on mortality and morbidity from breast cancer, research topics on breast cancer in men, topics related to the quality of life of breast cancer patients, breast cancer patient outcomes, genetic screening, genetic testing and also including gene expression profiling.

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## ETHICAL CONSIDERATIONS

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### Conflict of Interest Statement

No conflict interest in this research

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