REVIEW ARTICLE
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Prevalence and psychological repercussion of phantom breast syndrome in women undergoing mastectomy: a systematic review

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ABSTRACT

Breast cancer is the most common cancer among women in the world and the leading cause of death among Brazilian women. The presence of phantom breast syndrome (PBS) is one of the possible postoperative complications and may reach prevalences of up to 53% among mastectomized women. This study assessed the scientific evidence regarding the presence of PBS and its psychological repercussions in women undergoing mastectomy. This is a systematic review of observational studies based on the recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses methodology. The methodological quality of the studies and the level of scientific evidence were assessed using the Newcastle-Ottawa Scale and the Grading of Recommendations Assessment, Development and Evaluation. A total of 95 articles were identified, but only 11 met the eligibility criteria. The outcomes of the presence of PBS and psychological repercussion were evaluated in 2,160 and 1,996 patients, respectively. It was found that the prevalence of PBS varies according to age, being on average 28% and reaching up to 50% in women under 80 years of age. This phenomenon can occur from three months to six years after amputation, tending to regress over time. Anxiety, depression, and sleep disorders are the most prevalent psychological effects (35.8%, 31.5%, and 29.2%, respectively). The studies presented strong scientific evidence of PBS and moderate evidence of psychological repercussions associated with this context.

KEYWORDS: breast neoplasms; mastectomy; pain; phantom limb.

INTRODUCTION

Breast cancer is the most frequent type of cancer in women worldwide, being the leading cause of death for Brazilian women, with an estimated 66,280 new cases each year of the 2020–2022 period[1]. In 2017, 16,724 deaths from female breast cancer were recorded, equivalent to a risk of 16.2 deaths/100,000 women[1].

In recent years, advances have been made in the treatment of breast cancer, especially with regard to less mutilating surgeries[2]. Treatment varies according to the stage of the disease, and mastectomy is one of the available modalities.

The presence of phantom breast syndrome (PBS) is considered a postoperative complication. Although this was previously a rare complaint after mastectomy[3], recent studies have shown an increase in the frequency of PBS[4-6], suggesting that its true incidence and prevalence have been neglected in the past.

PBS is a painful sensorial experience of the removed breast, as if it were still present, characterized by "shocks" or "pins" in the most distal part of the breast (papilla)[1]. The pathophysiology of PBS can be understood as an overlapping of neighboring cortical zones, which can "invade" the representative territory of a nearby area or even by unmasking silent synapses[7]. PBS has a considerably variable prevalence estimate in the literature, reaching 53%, possibly reflecting differences in post-mastectomy follow-up time and factors such as anxiety, depression, and somatization. Listing the psychological disorders caused by PBS and knowing their prevalence will provide the basis for the elaboration of guidelines for prevention, diagnosis, and intervention related to this syndrome.

Given the above, the objective of the present study was to verify the scientific evidence regarding the presence of PBS and its psychological repercussions in women undergoing mastectomy.

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METHODS

A systematic review of observational studies was carried out, based on the recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)[8] methodology. The systematic review protocol was registered in the International Prospective Register of Systematic Reviews (PROSPERO), under number CRD42022347959.

The electronic search was carried out in the Scielo, Lilacs, and PubMed databases. The terms "breast", "phantom", "pain", and "syndrome", along with their correlates in English and Spanish, were established as keywords. Intentionally, broader terms were determined to enable the identification of a greater number of studies and, thus, minimize the risk that any relevant article could not be included in this survey. To cross the keywords, the logical operator "OR" was used.

To select the articles, the following inclusion criteria were considered:

1. texts published between 2002 and 2020;
2. in English, Portuguese, and Spanish; and
3. that addressed the topics of phantom breast, phantom breast syndrome, and phantom breast pain.

Duplicate articles were excluded due to simultaneous indexing in more than one database, whose analyzed population had previous comorbidity. Animal studies were also ruled out.

The presence of PBS and its psychological repercussions were considered as outcomes of interest to be reviewed by this study.

Bibliographic data were collected from November 2019 to July 2020, by two independent reviewers. The identified differences were resolved by consensus.

Based on the PRISMA[8] methodology, studies were identified and then evaluated using titles and abstracts. After excluding studies that did not meet the purpose of this systematic review, the articles were read in full and those considered unreliable (due to methodological issues or conflicts of interest) were disregarded. A specific form of data extraction was prepared by the authors, helping in the descriptive and critical analysis of the results, which were later grouped in a table in order to present the main information of the selected studies.

To assess the methodological quality of the studies, the Newcastle-Ottawa Scale (NOS)[9] was used, indicated to analyze evidence from observational studies. The selected articles were evaluated in three domains: sample recruitment and selection, similarity between participants and verification of the outcomes of interest.

The quality of scientific evidence was assessed using the Grading of Recommendations Assessment, Development and Evaluation (GRADE)[10] system and classified into four levels: high, moderate, low, and very low. Determining the level of evidence took into account the following factors: study design; limitations and methodological inconsistencies; indirect evidence; vagueness; publication bias; and residual confounders.

RESULTS

95 articles were identified, 58 of them located in scientific databases and 37 identified by cross-reference. After reading the titles and abstracts, 49 articles were excluded for not meeting the defined eligibility criteria. The NOS scale was applied to the remaining 46 studies, resulting in the exclusion of another 34. Therefore, 12 studies were selected for full reading. Then, another text was excluded because it also did not meet the eligibility criteria. Thus, the final sample consisted of 11 articles. Figure 1 shows the flowchart of the number of articles selected based on the inclusion and exclusion criteria.

After selecting the articles, the basic characteristics of the 11 included studies were listed in Table 1, facilitating the descriptive and critical analysis of the results obtained by the authors.

The evaluation of the quality of the evidence for the established outcomes is shown in Table 2. In the analysis using the GRADE system, a high degree of evidence was observed for PBS and moderate for psychological repercussions in women undergoing mastectomy.

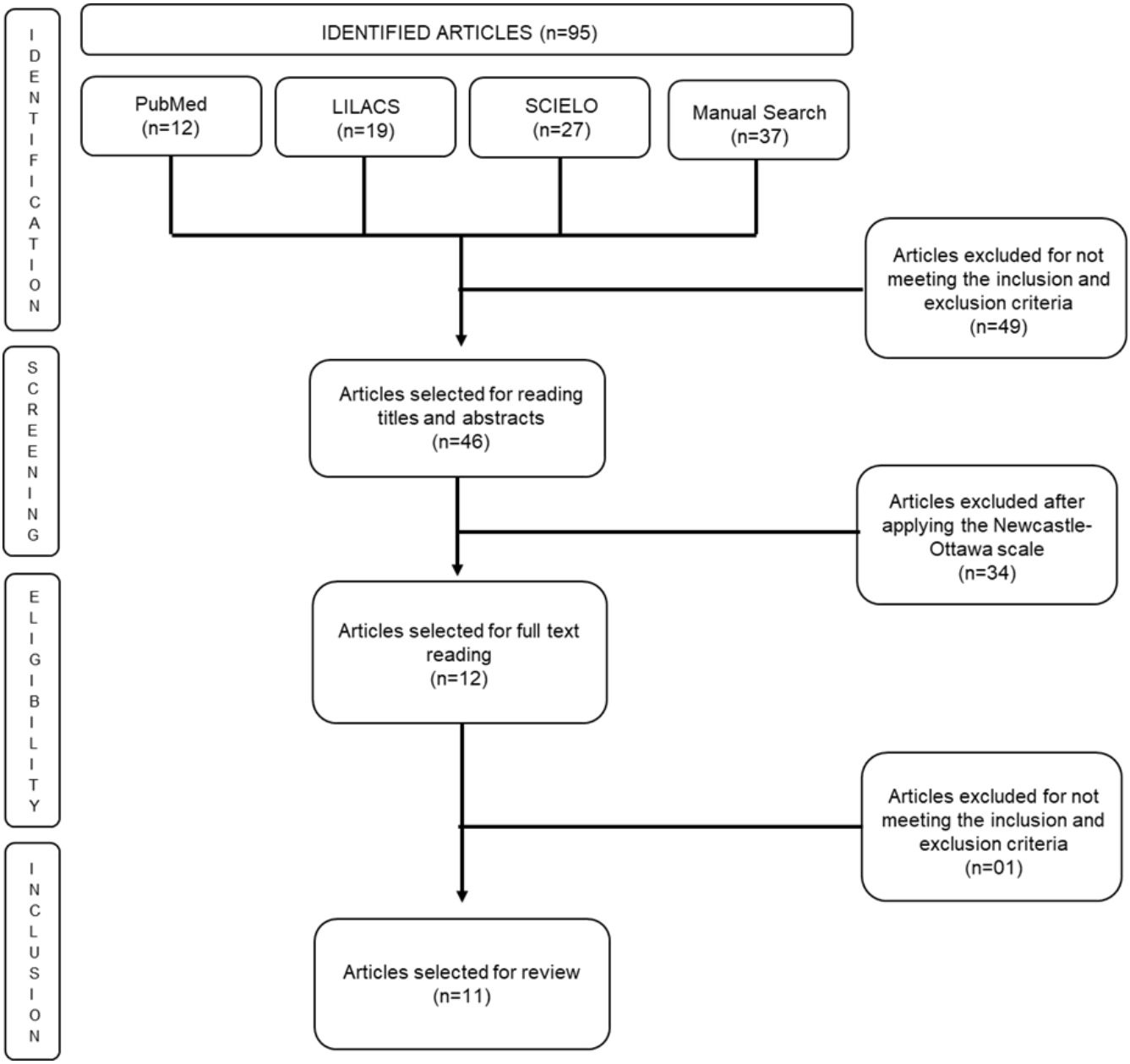
DISCUSSION

This review aimed to verify the scientific evidence regarding PBS and its psychological repercussions in women undergoing mastectomy. The analysis of the selected articles demonstrated the presence of PBS in all of them, with the development of some psychological disorder associated with pain in the phantom breast. It is noteworthy that all articles included had breast cancer as the cause of amputation, whose location and type of treatment employed were not related to the occurrence of the phenomenon[11].

Analyzing the prevalence of PBS, the study by Rothemund et al.[4], with 39 patients, recorded a prevalence of 23% of PBS among patients, in addition to its onset up to three months after the amputation. On the other hand, Dijkstra et al.[12], following up 204 patients for two years, demonstrated a PBS incidence of 13%. In addition, it was reported that, over time, the number of patients who reported PBS decreased, being of little clinical relevance in the 24 months after amputation[12].

Among the analyzed studies, the one by Bjorkman et al.[13] showed the highest prevalence of PBS (50%), but this research used a small sample (n=8), despite the recruitment having lasted six months. The study with the lowest prevalence (5.4%) concluded that, six weeks after the mastectomy, there was no anxiety caused by PBS, but the patients had significant depression and sleep disorders[14].

Spyropoulou et al.[15], evaluating women aged 59.4+11.4 years, demonstrated the same prevalence rate (6%) as in the study by Markopoulos et al.[16], who evaluated women aged 56.4+10.5 years, this being the lowest prevalence rate observed in the analyzed articles.



PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

Figure 1. Flowchart of article selection based on the PRISMA methodology.

In the study by Faria et al.[11], PBS was associated with the characteristics of painful sensation; five patients reported pain as mild or moderate, and two as unbearable pain. In addition, of the seven patients with PBS, five reported shock pain, one burning pain, and one pins and needles pain. Another study evaluating the pain scale revealed that about 75% of patients used weak opioids, non-steroidal anti-inflammatory drugs, or adjuvants to control pain[14].

Silva et al.[6] evidenced PBS aggravating and relieving factors, in which 63.6% of participants reported pain worsening in case of physical exertion and 90.9% improvement at rest. Ahmed et al.[14] demonstrated that pain was more frequent in the morning and increased when handwashing clothes, sitting down, lifting weights, in hot or cold environments, and under pressure and friction, recorded in one or two episodes per week.

Macdonald et al.[5] followed patients with PBS for a longer period (from three to nine years) and demonstrated that 17% of women will have post-mastectomy pain within 12 years. The prevalence of this study was cumulative to allow assessment of the variability of pain onset and remission.

As the data revealed, the main cause of mastectomy is breast cancer, which tends to grow from the age of 40[1]. The mean age of

Table 1. Summary of the main descriptive characteristics of the included studies.

Authors, year	Study design	Sample	Presence of PBS (%)	Time of onset of PBS (%)	Type of psychological disorder (%)	Follow-up of patients
Rothemund et al.[4]	Cross-sectional	39 post-BC women, aged 55.0+13.5 years	23	12 weeks	NI	NA
Macdonald et al.[5]	Longitudinal	175 post-BC women, aged 56.2+10.9 years	43	94 weeks	NI	36–108 months
Silva et al.[6]	Cross-sectional	98 post-BC women, aged 32 to 86 years (MA: 54 years)	30	Immediate: 10 12 weeks: 23.3 >12 weeks: 66.7	26.1 depression 27.2 anxiety	NA
Dijkstra et al.[11]	Longitudinal	204 post-BC women, aged 55.6+11.6 years	13	6 weeks	29.2 sleep disorder	24 months
Spyropoulou et al.[12]	Cross-sectional	105 post-BC women, aged 59.4+11.4 years	6	8 weeks	11 depression 6 anxiety 12.2 neuroticism	NA
Bjorkman et al.[13]	Longitudinal	8 post-BC women, aged 59.4+7.6 years	50	26–104 weeks	NI	24 months
Peuckmann et al.[14]	Cross-sectional	1.316 post-BC women, aged over 18 years <19	<19	NI	9 depression and sleep disorder	NA
Markopoulos et al.[15]	Cross-sectional	105 post-BC women, aged 56.4+10.5 years	6	NI	41 anxiety, depression, and sleep disorder	NA
Ahmed et al.[16]	Longitudinal	80 post-BC women, aged 49.3+12.7 years	5.4 (6 weeks) 8.2 (6 months) 13.6 (12 months)	6 weeks	80 depression 100 sleep disorder	24 months
Medina et al.[17]	Longitudinal	88 post-BC women aged <60 years ***vs.*** ≥60 years	<10	6.5 weeks: 44.3 26 weeks: 34.9 104 weeks: 18.2	68.8 anxiety, depression, and sleep disorder	9 months
Faria et al.[18]	Cross-sectional	40 post-BC women, aged 30 to 60 years	17.5	90 weeks	NI	NA

BC: breast cancer; NI: not informed; NA: not applicable; PBS: phantom breast syndrome.

Table 2. Quality of evidence of the assessed outcomes.

Outcome	Total number of study participants	Quality of evidence (GRADE)
Presence of PBS	2.160	High
Psychological repercussion	1.996	Moderate

PBS: phantom breast syndrome; GRADE: Grading of Recommendations Assessment, Development and Evaluation.

women undergoing radical mastectomy is 62.9 years[17], with PBS being one of the most frequently reported postoperative complications. It was possible to verify, in this systematic review, that PBS has a variable prevalence according to the age of the patients, observing the average prevalence of 28%, with a minimum of 6% and a maximum of 50% for women up to 80 years old, and mean prevalence of 18%, with a minimum of 6% and a maximum of 30% for women aged up to 86 years. There is, therefore, an important variability in the prevalence of PBS in the literature. Therefore, the adoption of a standard criterion for pain symptoms can be of great value, as it will allow a better comparative analysis of different studies.

Furthermore, PBS compromises people's daily activities, causing psychological disorders. Among the 11 studies analyzed, seven[6,12,14-16,18,19] demonstrated the influence of PBS on psychic well-being. Anxiety, depression, and sleep disturbance were the most common psychological disorders. As reported by Yurek et al.[20], depression was associated with younger patients with PBS as the breast has an important aesthetic and sexual significance for these women.

Regarding the psychological profile, depression and anxiety were the most relevant findings, commonly associated with PBS. The highest percentage of depression cases found in cross-sectional studies was 26.1%, of which 66.7% appeared three months after PBS[6]. The time of onset of symptoms fluctuated a lot, from immediately to two years after radical mastectomy. Of the cross-sectional studies that demonstrated the presence of anxiety, the article by Markopoulos et al.[16] was the one with the highest prevalence (41%).

Medina et al.[19] found that 68.8% of participants had changes such as anxiety, depression, and sleep disturbance after breast removal, but did not find an association between mood swings and PBS. Fakhari et al.[21], on the other hand, suggest that these alterations have a negative impact on the patients' quality of life,

resulting in non-adherent treatments and, finally, leading to a poor prognosis. It is worth mentioning that all studies evaluated in this systematic review considered only psychological disorders developed after amputation. Furthermore, Medina et al.[19] highlighted the importance of psychological support, with guidance related to bodily changes, support from the spouse and preoperative guidance, thus minimizing the patients' emotional manifestations.

Among the analyzed studies, Ahmed et al.[14] showed the highest prevalence of psychiatric disorders, with 100% of patients presenting sleep disorders. It is important to point out that all the analyzed studies referred to sleep disorders but did not specify them. Another relevant finding was the gradual increase in depression from 20.5% in six weeks to 56% in one year[14]. Patients also showed greater use of anxiolytics[14]. Silva et al.[6] recorded 26.1% of depression, 27.2% of anxiety, and 32.6% did not present any specific symptoms. Although there was a balance between patients with or without symptoms, Silva et al.[6] reported their exacerbation.

It is possible that depression and anxiety accompany most mastectomized women, as the breast is a symbol of femininity, and its loss causes feelings such as shame, rejection, and guilt[15]. Furthermore, Spyropoulou et al.[15] suggest that, currently, it is difficult to discern whether PBS leads to depressive symptoms or the depressive state is the predisposing factor for PBS.

CONCLUSION

This systematic review demonstrated strong scientific evidence of PBS in women undergoing mastectomy and moderate psychological repercussions associated with this context. It is important to emphasize that the prevalence of PBS varies according to the age of the patients, with a mean prevalence of 28%, which can reach up to 50% in women under 80 years of age. This phenomenon can occur from three months to six years after the amputation, tending to regress with time. Furthermore, the studies revealed that anxiety, depression, and sleep disturbance were the psychological disorders most commonly presented by women with mastectomies.

AUTHORS' CONTRIBUTIONS

JVLSN: Conceptualization, Data curation, Investigation, Writing – original draft, Writing – review & editing. MMS: Conceptualization, Data curation, Investigation, Writing – original draft, Writing – review & editing. KMA: Conceptualization, Data curation, Formal Analysis, Methodology, Validation, Writing – original draft, Writing – review & editing. CCPFM: Conceptualization, Data curation, Methodology, Project administration, Supervision, Validation, Writing – original draft, Writing – review & editing.

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