ABSTRACT

Background: Phantom Breast Syndrome (PBS) is one of the complications of mastectomy and it is type of condition in which patients have a sensation of residual breast tissue and can include both non-painful sensations as well as phantom breast pain. The sensation in PBS is different than a pain before mastectomy or a sensation related with scar tissue; the sensation in the removed breast is a neuropathic pain. PBS was divided into a sub-types with painful phantom and non-painful phantom sensations.

Objective: To review the literature to define the prevalence and clinical features of phantom breast syndrome.

Methodology: The studies conducted between 2004-2014 were reviewed through Medline Complete, Pubmed, Science Direct, and EBSCO and 11 studies were included in the present review.

Findings: In the samples of these 11 studies; there were 627 patients who had mastectomy and 7.8% of them (49 patients) were diagnosed with phantom breast syndrome, 65.8% (413 patients) had phantom breast sensation and 6.7% (42 patients) had phantom breast pain.

Conclusion: It is needed to define certain standards in the assessment of PBS, professional meetings should be done for the treatment and care of patients with PBS. In addition, more randomized controlled studies are needed in this subject.

Key words: Phantom breast syndrome, phantom breast, phantom pain, phantom sensation, mastectomy
Phantom breast pain can be seen with phantom breast sensation and this situation is called phantom breast syndrome (PBS). PBS is one of the complications of mastectomy, which is one of the main surgical treatment options of breast cancer. The sensation of PBS may occur with feeling the whole or half of the removed breast or only the nipple. The type of the sensation can be described as itchiness, heaviness or tingling and these sensations can be with or without pain. Generally the phantom breast is felt exactly in the previous size and shape, however; certain patients may feel their phantom breast heavier and bigger than before. The pathophysiology of PBS is not well known yet, but it may be seen in 7-17.4% of the patients who had mastectomy (1,2,3).

Methodology

Literature research was done with the keywords “phantom” and “mastectomy” in EBSCHO, PubMed, Medline Complete, and Science Direct databases. In total 1383 studies were found. The research results are restricted with the studies that can be downloaded as full-text, published in the last 10 years, written in Turkish and English. After the restrictions, the total number of studies found was decreased to 713. After the evaluation of the titles and abstract of these studies; 18 studies in total were reviewed as full-texts. According to the Qualitative Assessment and Review Instrument (QARI) of Joanna Briggs Institute, retrospective descriptive, prospective descriptive, cohort and cross-sectional 11 studies were included in the review. The 7 studies were excluded because of insufficient date related to PBS, inclusion of same sample with a similar study, and qualitative study design. Grey literature research was done in National Thesis Center of Turkey, Thesis Center of Canada, and website of System for Information Grey Literature in Europe, database of ProQuest Dissertations & Theses Global with the keywords “phantom” and “mastectomy”. Two theses were reached in the database of ProQuest Dissertations & Theses Global and these two studies were included in the review. Consequently, the present review included 11 studies total which included all PBS patients after mastectomy, reached in full-text in Turkish/English. Evaluation process and exclusion reasons were explained in Figure 1. The studies were evaluated according to their methodology, the year published, sample size and features, incidence of phantom breast pain, and phantom breast sensation, and clinical features.

The studies included in the present review were evaluated by the Qualitative Assessment and Review Instrument (QARI) of Joanna Briggs Institute according to their methodology, objectives, data analysis, sample characteristics; and the ones that achieve at least 8 criteria out of 10 were included. The criteria of the Qualitative Assessment and Review Instrument (QARI) of Joanna Briggs Institute were as follows:

1. There is congruity between the stated philosophical perspective and the research methodology.
2. There is congruity between the research methodology and the research question or objectives.
3. There is congruity between the research methodology and the methods used to collect data.
4. There is congruity between the research methodology and the representation and analysis of data.
5. There is congruity between the research methodology and the interpretation of results.
6. There is a statement locating the researcher culturally or theoretically.
7. The influence of the researcher on the research, and vice versa, is addressed.

8. Participants, and their voices, are adequately represented.

9. The research is ethical according to current criteria or, for recent studies; there is evidence of ethical approval by an appropriate body.

10. Conclusions drawn in the research report do appear to flow from the analysis, or interpretation, of the data.

Findings

In the present review, 11 studies published between 2004-2014 and done with women who have undergone mastectomy and experienced phantom breast syndrome, phantom breast pain, and phantom breast sensation; their prevalence and clinical features. The literature research process is explained in Figure 1.

The studies of last 10 years that provide information related to phantom breast pain and sensation after mastectomy and their clinical features included in the present review. The studies excluded were a qualitative study that doesn’t provide sufficient data, a review study, a study that has the same sample characteristics with a similar study, and 4 studies that don’t provide enough data about phantom breast.

According to the data of the reviewed studies;

Features of the Reviewed Studies:
The Sample:
The age distribution of the sample was between 18-80 years.

The samples of the reviewed studies included women who have undergone breast surgery including mastectomy. The smallest sample size was 28 (4), and the biggest sample size was 1131. The total number of all the samples was found 2514.

Among the investigated studies; 627 patients have had mastectomy, 1074 patients had Axillary Lymph Node Dissection (ALND), 578 patients had Sentinel Lymph Node Dissection (SLND), and 63 patients had reconstructive surgery.

The total number of the patients who have received chemotherapy was 904, while the ones who have received radiotherapy were 1083.

Methods

The studies included in the present review consisted of 4 prospective-descriptive studies, 4 retrospective-descriptive studies, 1 prospective-cohort study, 1 cross-sectional study and 1 qualitative study. The data collection methods were face to face interview, phone interview, and interview with e-mail communication.

Data Collection Tools Used

For the evaluation of the pain 0-10 or 0-100 Visual Analog Scales have been used. The occurrence of phantom breast sensation has been evaluated with "BSAS (Breast Sensation Assessment Scale)". The parameters related to the quality of life have been assessed by (EORTC QLQ C30 and BR-23), CARS (Dutch version of Concerns about Recurrence Scale), and DASS (The Depression Anxiety Stress Scale-21). Activity and functionality related features have been evaluated by MRC (Medical Research Council Scale), and FIM (Functional Independence Measure).

Results of the Reviewed Studies

The studies were reviewed in terms of phantom breast syndrome, phantom breast sensation, and phantom breast pain; and their prevalence, frequency, localization, the level
of distress, accompanying symptoms, and affecting factors.

Prevalence of Phantom Breast Syndrome, Phantom Breast Sensation, and Phantom Breast Pain:

One study reported that phantom breast syndrome was found in 9 patients (23%) out of 39 who had mastectomy (5). In another study, phantom breast syndrome was diagnosed among the 24 women (22.8%) who had breast surgery out of 105 (6). Another study reported phantom breast syndrome in 16 patients (57%) out of 28 who had breast surgery (4). In the grey literature research, in the dissertation study, phantom breast syndrome was found in 40% of the sample (7).

During the patient evaluations between 24th–60th months after breast surgery; one study reported 23 patients who keeps having phantom breast sensation (16). In another study, the number of women with phantom breast sensation was reported as 15 (8). According to a semi-experimental study; 34% of the women (n=128) stated to have phantom breast sensations within the 1st month after mastectomy (9). One study investigated 1131 women who had mastectomy and were found the prevalence of phantom breast sensation as 25% (10). Some other study result one by one; the prevalence of phantom breast sensation was found 15% among 85 patients (11), 19% among 74 patients (12), 9.5% among 80 patients in the 12th month and this ratio was increased to 17% (13). One another study reported that 21 patients out of 28 stated phantom breast sensation after breast surgery (4).

One study investigated 174 women who had mastectomy and reported that on the 50th day after mastectomy, the prevalence of phantom breast pain was found 6.1% (14). In the same study sample; 8 months after mastectomy, phantom breast pain was seen 3.2% of the sample. In another study, among 85 patients, 5.6% stated phantom breast pain (11). Some other study result one by one; 1% phantom breast pain among 74 patients (12), 5.4% among 80 patients (13). %4 among 317 patients (15). In another study was found that among 28 patients, 17 of them reported phantom breast sensation (4).

The Frequency of Phantom Breast Sensation and Pain:

One study reported 3 women started that they experience phantom breast sensation once a week, 5 women said once a month, 3 women said once a month or less. In the same study, regarding phantom breast pain; 2 women reported to have phantom breast pain couple of times a week, 2 women said once a week, and 1 women said couple of times a day (5).

One study reported that in their sample, 9 women stated to have phantom breast sensation couple of times a year, 3 women said couple of times a month, 2 women said that they have constant phantom breast sensation while only 1 women stated to have phantom breast pain couple of times a year (12).

In a study reported 9.1% of the sample were stated to have phantom breast sensation once a day, 22.7% answered as once a week, while 13.6% said at least once a month (6).

One study reported that the frequency of phantom breast pain was 1-2 times in 10 patients, while 7 patients reported that they have phantom breast pain couple of times a day (13).

According to the grey literature, 137 women who had mastectomy and reported that phantom breast sensations are starting following 1-6 months after surgery and this sensation appears frequently during the day especially while resting (7).

Location and Severity of Pain and Sensation, and their Level of Distress:

One study reported 4 patients have phantom breast pain on the nipple, 4 stated to
have pain in all the phantom breast, 1 patient reported pain in the left lower quadrant while 1 other patient reported pain in the left upper quadrant of the phantom breast (5). The average magnitude of non-painful phantom sensations was 40.0 (SD: 13.6; Range: 20-60). The minimum was 25.7 (SD: 17.5; Range: 0-50) while the mean maximum was 51.0 (SD: 17.8; Range: 20-80). In the same study, 3 patients stated phantom breast pain in the phantom nipple, 4 patients stated pain in the whole breast, 1 patient said the pain is on the left upper quadrant, 1 patient reported pain both on the left upper quadrant and the nipple, while 1 patient stated pain on the right upper quadrant of the phantom breast. The characteristics of these reported painful sensations were described as twinging, tearing, cutting, tense, sharp, convulsive, pressing, and cramp-like. The average amount of phantom pain was 60.0 on VAS (SD: 16.6; Range:40-90) (5).

One study stated among the patients who had phantom breast sensation, 50% stated that the sensation includes all the breast, while 29.2% said the sensation is localized with the nipple, and 20.8% said that the sensation is limited in a certain part of the phantom breast. Non painful phantom sensations were described as itching, experienced by 54.5% of PBS patients, pinpricks reported by 70.8%, pins and needles by 16.7% and sense of heaviness by 8.3%. Phantom pain was experienced by six patients (25% of PBS patients), who concurrently reported non painful phantom sensations (itching, pinpricks and pins and needles) at the phantom breast.

PBS was localized in the entire breast in 50% of PBS patients, only in the nipple in 29.2% and concerned only a part of the breast in 20.8%. In the majority of PBS cases, phantom experience had the size (88.9%) shape (76.5) and weight (64.7%) of the normal breast. In this study, the satisfaction level with the phantom breast sensation was reported as 13% (10).

One study reported that their study sample stated that the phantom breast sensation/pain was vivid and the breast was felt like its old own size and shape. Six patients out of 17 described the phantom breast pain as agonizing on the verge of unbearable. Among 21 patients who were experiencing phantom breast sensation, 6 patients defined this sensation as mild, while 6 other patients explained it as modest and manageable. Among 17 patients with phantom breast pain, 7 patients described the pain as inscrutable but not frightening. On the other hand, 8 patients with phantom breast sensations in the sample described this sensation as nasty and gnawing, while some other 8 patients defined it as inscrutable but not frightening (4).

According to one study, study sample who have phantom breast pain, described the pain as an unpleasant sensation of breast presence like pin-prick, burning or torsion (14). In another study stated that the patients with phantom breast pain defined it as sharp, aching, and dull (11).

According to one study, 80% of the women with phantom breast pain stated that the pain is in the entire breast; while 40% said the pain is in the nipple area in moderate level. In this study, VAS score for phantom breast pain was found 5.25, 4.8, and 5.4 at 6 weeks, 6 and 12 months respectively (13).

One study investigated 22 patients having phantom breast sensation after SLND and ALND surgery. Two of the sample explained this situation as very severe and very much distress. Seventy percent of the sample stated that the sensation doesn’t affect their quality of life. Thirty percent of SLNB patients and 26% of ALND patients stated that they experienced at least one quality of life problem. Fear of recurrence, followed by problems related to body image, were the
most frequently reported quality of life problems in both groups (8).

In another study, women who were experiencing phantom breast sensation stated that as it came and went, and the levels of severity and distress were low. In the same study, it was found that the body image and recurrence scores were high in the quality of life scale (16).

According to one study the women stated their phantom breast pain as little, and the phantom breast sensation as moderate. In the same study, moderate VAS score of phantom breast pain was found between 0.2-0.6 (mean 0.4±1.2). Regarding the quality of life, the researchers stated that role function, insomnia, hair loss, body image scores were found high in the quality of life scale among the women with phantom breast pain and sensation (12).

In the grey literature, one study shows that the characteristic of phantom breast sensation was defined by patients as itching, mild pain, and pressure on the nipple of the phantom breast. The most common definition was found “pulling sensation” (9).

Accompanying Symptoms and Affecting Factors:

In one study a significant relationship were found between depression, sleep disturbance and anxiolytic usage among 80 patients after mastectomy. In the same study, among the women with phantom breast pain; in the 6th week, 6th month, and 12th month follow-ups, frequency of depression was found 50%, 100%, and 80% (p<0.05); frequency of sleep disturbance was found 50%, 83.3%, and 100% (p<0.05) respectively (13).

When the study investigated (13) the factors that increase or reduce the phantom breast pain; they have found that mornings times (4 patients), friction of the clothes (10 patients), sitting (8 patients), exercising (4 patients), lifting heavy objects (7 patients), lying down on the affected side (1 patients), and taking a shower (1 patient) were found as the factors increasing the phantom breast pain. In addition, it was found that resting (5 patients), exercising (6 patients), putting pressure (4 patients), feeling cold (7 patients), doing frictions (5 patients) were the factors that reducing the severity of phantom breast pain. In the same study, it was also found that phantom breast sensation were found more frequently in the women who works regularly, and among the woman with phantom breast sensation; depression, sleep disturbances and anxiolytic usage were more common (p<0.05) (13).

A study reported (5) cancer treatment wasn’t found significantly related to phantom breast syndrome whether painful or not. Besides, reconstructive surgery was also not related to phantom breast syndrome.

A study was found significant association between PBS and age (10), and reported that PBS was more common among young women (OR 1.030 per year; 95% confidence interval (CI) 1.010-1.050; p=0.0026). Pain in the breast area was found associated with an increased prevalence of PBS (OR 2.999; 95% CI 2.251-3.997; p<0.0001). The woman who received ALND surgery had a significantly lower prevalence of PBS than those receiving SLND surgery (OR 0.645; 95% CI 0.420-0.991; p=0.0456). In addition; neither chemotherapy (OR 0.909; 95% CI 0.638-1.295; p=0.5970) nor radiotherapy (OR 0.907; 95% CI 0.630-1.307; p=0.6013) was associated with PBS.

In another study (14) the body mass index of the patients who had phantom breast sensation was found 28±5 in average and 72.3% of the samples were in obese classification.

A study (12) was found that none of the treatment and pre-mastectomy risk factors related factors was significantly associated with PB sensation or PB pain.
In the grey literature, when the patients are more stressed, nervous, and angry; they stated to feel the phantom breast syndrome more (7).

A study (13) was found that anxiolytic usage was significantly higher among the woman with phantom breast pain (p<0.05). Three-quarters of the women with phantom breast pain were managing their pain with weak opioids, NSAIDs and adjuvant medicines. Among the women with phantom breast pain, 25% of them were using morphine in 6 weeks, while 20% said in 12 months. The women who were using NSAIDs were found as 50% (6 weeks), 33% (6 month), and 30% (12 months) respectively. Tramadol usage was 75% (6 weeks), 83% (6 months), and 60% (12 months); while gabapentin usage was 5% (6 weeks), 66% (6 months), and 60% (12 months) (13).

The studies investigated in this literature review have been listed in Table 1 and 2.

### Table 1. Descriptive Features of the Investigated Studies in this Review

<table>
<thead>
<tr>
<th>Author(s) and Year</th>
<th>Sample Size</th>
<th>Age Distribution</th>
<th>Design of the Study</th>
<th>Methodology</th>
<th>The period that the study is followed</th>
<th>Type of Surgery (n)</th>
<th>Chemotherapy (%)(y)</th>
<th>Radiotherapy (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Badin et al. 2004</td>
<td>204</td>
<td>27-85</td>
<td>Prospective, descriptive</td>
<td>Face-to-face interview</td>
<td>3-24 months</td>
<td>Total Mastectomy</td>
<td>ALND</td>
<td>SLND</td>
</tr>
<tr>
<td>Rothermund et al. 2004</td>
<td>30</td>
<td>55.1-11.6 (12-77)</td>
<td>Retrospective, descriptive</td>
<td>Phone calls</td>
<td>6-204 months</td>
<td>-</td>
<td>39</td>
<td>39</td>
</tr>
<tr>
<td>Badin et al. 2007</td>
<td>187</td>
<td>34-84</td>
<td>Retrospective, descriptive</td>
<td>Face-to-face interview, e-mail</td>
<td>0-60 months</td>
<td>64</td>
<td>17</td>
<td>-</td>
</tr>
<tr>
<td>Dijkstra et al. 2007</td>
<td>74</td>
<td>55.6±12.8</td>
<td>Prospective, descriptive</td>
<td>Face-to-face interview, medical records</td>
<td>0-24 months</td>
<td>74</td>
<td>19</td>
<td>-</td>
</tr>
<tr>
<td>Steegers et al. 2008</td>
<td>317 *Med 60</td>
<td>Retrospective, descriptive</td>
<td>Face-to-face interview, phone calls</td>
<td>0-6 months</td>
<td>167</td>
<td>105</td>
<td>-</td>
<td></td>
</tr>
<tr>
<td>Markopoulos et al. 2010</td>
<td>108</td>
<td>59.6±11.4 (30-86)</td>
<td>Cross-sectional</td>
<td>Face-to-face interview, e-mail</td>
<td>1-6 months</td>
<td>-</td>
<td>50</td>
<td>-</td>
</tr>
<tr>
<td>Hansen et al. 2011</td>
<td>1131</td>
<td>18-70</td>
<td>Retrospective, descriptive</td>
<td>-</td>
<td>850</td>
<td>231</td>
<td>-</td>
<td></td>
</tr>
<tr>
<td>Khosh et al. 2012</td>
<td>85</td>
<td>Med 57 (33-80)</td>
<td>Retrospective, descriptive</td>
<td>-</td>
<td>1-24 months</td>
<td>14</td>
<td>25</td>
<td></td>
</tr>
<tr>
<td>Bjoekman et al. 2012</td>
<td>28</td>
<td>Med 57</td>
<td>Retrospective, descriptive</td>
<td>-</td>
<td>14</td>
<td>25</td>
<td>63</td>
<td>63</td>
</tr>
<tr>
<td>Nogueira et al. 2012</td>
<td>174</td>
<td>58±13</td>
<td>Prospective cohort</td>
<td>Medical records</td>
<td>6 months</td>
<td>127</td>
<td>-</td>
<td>134</td>
</tr>
<tr>
<td>Ahmed et al. 2014</td>
<td>80</td>
<td>49.3±12.7 (26-71)</td>
<td>Prospective, descriptive</td>
<td>Face-to-face interview</td>
<td>6 weeks-12 months</td>
<td>80</td>
<td>-</td>
<td>36</td>
</tr>
</tbody>
</table>

*Std. Dev.: Standard Deviation  
*Med. Median

### Discussion

Among the studies investigated in this literature review; the prevalence of Phantom Breast Syndrome was 23% (5) 22.9% (6) and 57% (4). In the literature; this ratio change as 35% (17), 29.8% (18), 23.3% (19), %25.8 in three weeks after mastectomy (20), 13.6% in one month after mastectomy (21), 24.5% after a year (20). According to these findings, it can be said that Phantom Breast Syndrome is seen more frequently in the last 10 years.

Phantom breast sensation was seen in the 38% (16), 51% (5), 48% (8), 19% (12), 26% (10), 15% (11) and respectively 9.5% in the 6th week, 6.8% in the 6th month and 17% in the 12th month in the sample of another study (13). In the literature, the ratios for the prevalence of phantom breast sensation were as follows:26% (22), 60% (23), %40 (24) %33,5 (25) and %34 (26),15% 3 weeks after mastectomy, 11.8% 1 year after mastectomy, and 11.8% 6 years after mastectomy (27).Prevalence of phantom breast sensation in the studies investigated in this review is found similar to the findings in the early literature.

Prevalence of phantom breast pain in the studies investigated in this review were as follows: 1% (12), 3.7% (15), 6% (11), 6.1% (14), 5.4% in the 6th week after mastectomy, 8.2% in the 6th month after mastectomy, 13.6% one year after mastectomy (13). In the early literature, the prevalence rates were 13.3% three weeks after mastectomy, 12.7% one year after mastectomy (20), and 17.4% six years after mastectomy (28), 13.3% 3 weeks after mastectomy, 12.7% 1 year after mastectomy, and 17.4% 6 year after...
According to these ratios, it can be said that the prevalence of phantom breast pain in the last decade is parallel with the findings in the literature.
In the literature, it is stated that phantom breast syndrome is mostly seen among women <60 years old (22, 23, 24, 25) and in this review, it was found that phantom breast syndrome is frequently seen among women younger than 40 years old (10, 13). In some studies reported that phantom breast sensation is mostly seen in the nipple and the whole breast (18,23,25). In this systematic review, the investigated studies were in parallel with these findings and phantom breast sensation was mostly seen in the nipple and in the whole breast (5, 6, 13).

In the investigated studies; there wasn’t any significant relationship between chemotherapy/radiotherapy and phantom breast sensation (17, 20, 28).

**Conclusion**

The prevalence of phantom breast syndrome, phantom breast pain and phantom breast sensation were found as 49 patients with phantom breast syndrome (7.8%), 413 patients with phantom breast sensation (65.8%) and 42 patients with phantom breast pain (6.7%) in total of 627 patients. The occurrence rates of phantom breast sensation/pain were as the first 15 days after surgery up to 60 months. The age distribution was 20-85 and phantom breast syndrome was more frequently seen in patients younger than 40 years old. The localization of phantom breast syndrome was more frequently in the whole breast. It was seen that phantom breast sensation and pain is increasing the depression level, decreasing the quality of life, causing sleep disturbances, increasing anxiolytic usage and affecting body image negatively.
Phantom breast syndrome is one of the clinical problems after mastectomy. To be able to define phantom breast syndrome correctly, a reliable and valid scale that consists of questions specific to this syndrome. Besides, more studies that are multicenter and only consisted of patients with phantom breast syndrome are needed to be able to define the variables affecting phantom breast syndrome.

Limitations

This systematic review was done with 11 studies. In these studies the evaluation periods of phantom breast syndrome, phantom breast sensation and phantom breast pain were different in each study. Therefore it wasn’t possible to use a common classification to define the frequency of symptoms related to phantom breast syndrome/sensation/pain.

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References

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