



# “It still affects our economic situation”: long-term economic burden of breast cancer and lymphedema

Lorraine T. Dean<sup>1,2</sup>  · Shadiya L. Moss<sup>3</sup> · Yusuf Ransome<sup>4</sup> · Livia Frasso-Jaramillo<sup>5</sup> · Yuehan Zhang<sup>6</sup> · Kala Visvanathan<sup>1,2</sup> · Lauren Hersch Nicholas<sup>2,5</sup> · Kathryn H. Schmitz<sup>7</sup>

Received: 20 November 2017 / Accepted: 9 August 2018 / Published online: 18 August 2018  
© The Author(s) 2018

## Abstract

**Purpose** Financial toxicity after breast cancer may be exacerbated by adverse treatment effects, like breast cancer-related lymphedema. As the first study of long-term out-of-pocket costs for breast cancer survivors in the USA with lymphedema, this mixed methods study compares out-of-pocket costs for breast cancer survivors with and without lymphedema.

**Methods** In 2015, 129 breast cancer survivors from Pennsylvania and New Jersey completed surveys on demographics, economically burdensome events since cancer diagnosis, cancer treatment factors, insurance, and comorbidities; and prospective monthly out-of-pocket cost diaries over 12 months. Forty participants completed in-person semi-structured interviews. GLM regression predicted annual dollar amount estimates.

**Results** 46.5% of participants had lymphedema. Mean age was 63 years (SD = 8). Average time since cancer diagnosis was 12 years (SD = 5). Over 98% had insurance. Annual adjusted health-related out-of-pocket costs excluding productivity losses totaled \$2306 compared to \$1090 ( $p = 0.006$ ) for those without lymphedema, or including productivity losses, \$3325 compared to \$2792 ( $p = 0.55$ ). Interviews suggested that the cascading nature of economic burden on long-term savings and work opportunities, and insufficiency of insurance to cover lymphedema-related needs drove cost differences. Higher costs delayed retirement, reduced employment, and increased inability to access lymphedema care.

**Conclusions** Long-term cancer survivors with lymphedema may face up to 112% higher out-of-pocket costs than those without lymphedema, which influences lymphedema management, and has lasting impact on savings and productivity. Findings reinforce the need for actions at policy, provider, and individual patient levels, to reduce lymphedema costs. Future work should explore patient-driven recommendations to reduce economic burden after cancer.

**Keywords** Breast cancer · Lymphedema · Financial toxicity · Economic burden

**Electronic supplementary material** The online version of this article (<https://doi.org/10.1007/s00520-018-4418-4>) contains supplementary material, which is available to authorized users.

✉ Lorraine T. Dean  
ldean9@jhu.edu

<sup>1</sup> Department of Epidemiology, Johns Hopkins Bloomberg School of Public Health, Johns Hopkins University, 615 N Wolfe St, E6650, Baltimore, MD 21205, USA

<sup>2</sup> Department of Oncology, Johns Hopkins School of Medicine, Baltimore, MD, USA

<sup>3</sup> Department of Epidemiology, Mailman School of Public Health, Columbia University, New York, NY, USA

<sup>4</sup> Department of Social & Behavioral Sciences, Yale School of Public Health, New Haven, CT, USA

<sup>5</sup> Department of Health Policy & Management, Johns Hopkins Bloomberg School of Public Health, Johns Hopkins University, Baltimore, MD, USA

<sup>6</sup> Department of General Internal Medicine, University of Pennsylvania Perelman School of Medicine, University of Pennsylvania, Philadelphia, PA, USA

<sup>7</sup> Department of Public Health Sciences, Pennsylvania State University College of Medicine, Pennsylvania State University, Hershey, PA, USA

## Introduction

Nearly half of cancer survivors experience financial distress [1]. Cancer-related financial toxicity, the harmful personal economic burden caused by cancer treatment [2], affects nearly half of cancer survivors [1] and is present even among those with health insurance [2–6]. Costs of cancer care are even higher for those with adverse treatment effects [7–12], such as breast cancer-related lymphedema [13, 14], and comorbidities [3, 15].

Breast cancer-related lymphedema affects up to 35% [16, 17] of the 3.5 million breast cancer survivors in the USA (2016) [18]. Breast cancer-related lymphedema is a chronic inflammatory condition that arises when there is disruption of lymphatic flow due to surgery, adjuvant radiation, and some forms of chemotherapy, infection, obesity, or other trauma to the lymphatic system [17, 19], leading to buildup of lymphatic fluid in the upper body, especially the arms, breast, and torso [20, 21]. The arm swelling and altered lymphatic function caused by lymphedema may affect a breast cancer survivor's ability to complete activities of daily living and maintain employment, leading to psychosocial distress, secondary comorbidities [19, 22–24], and limited work and career opportunities [25–27].

Previous work has estimated that incremental costs due to lymphedema for US cancer survivors at \$14,877 (excluding cancer-related costs) in the first 2 years after cancer treatment initiation [13]; however, these estimates are nearly 15 years old, focused on only the short-term costs, and predate the 2010 Affordable Care Act that expanded coverage for cancer-related care and banned refusal of coverage for those who might have a pre-existing condition [28]. A patient's decision about whether or not to expend resources on medical care versus other competing needs is driven by out-of-pocket costs. Yet, previous estimates rely on claims and administrative data, which neglect the impact of out-of-pocket costs. Altogether, data on out-of-pocket costs of lymphedema management is lacking for US-based samples and overlooks the long-term impact of cost and indirect costs, such as lost productivity.

To address gaps in this research topic and expand on previous inquiry, we conducted a prospective longitudinal explanatory mixed methods study. This is the first study in the USA to quantitatively compare long-term out-of-pocket direct and indirect costs among women with breast cancer-related lymphedema to those without a lymphedema diagnosis, integrating qualitative data to offer insight into what makes costs different comparing those with or without lymphedema.

## Methods

### Sample

From May to September of 2015, 258 women were screened by phone for eligibility to the PAL Social Economic and

Quality of Life (PAL SEQL) follow-up study. Recruits were identified from prior participants of the Physical Activity and Lymphedema (PAL) trial ( $n = 295$ ) [29, 30] who were still alive or participants who were ineligible ( $n = 163$ ) for the ongoing Women in Steady Exercise Research (WISER) Survivor Study ([ClinicalTrials.gov #NCT01515124](https://clinicaltrials.gov/ct2/show/study/NCT01515124)) [31], but met requirements for entry into PAL. All participants who had agreed to be contacted about future studies and had up-to-date contact information were contacted. Eligibility criteria included women with stages I–III invasive breast cancer, completion of active breast cancer treatment, > 1 lymph node removed, and current residents of Pennsylvania or New Jersey. Those with active cancer, or who were pregnant or planning to become pregnant in the next 6 months were excluded. Of those screened, 37 were ineligible, and 96 declined or dropped out due to lack of time to commit to a longitudinal study. Figure 1 describes the conceptual overview of the mixed methods study design.

### Ethical considerations and informed consent

The Institutional Review Board of the University of Pennsylvania approved the study. Written informed consent was obtained from all individual participants. Participants completed a measurement visit at baseline and 6 months that included measures of upper extremity lymphedema severity.

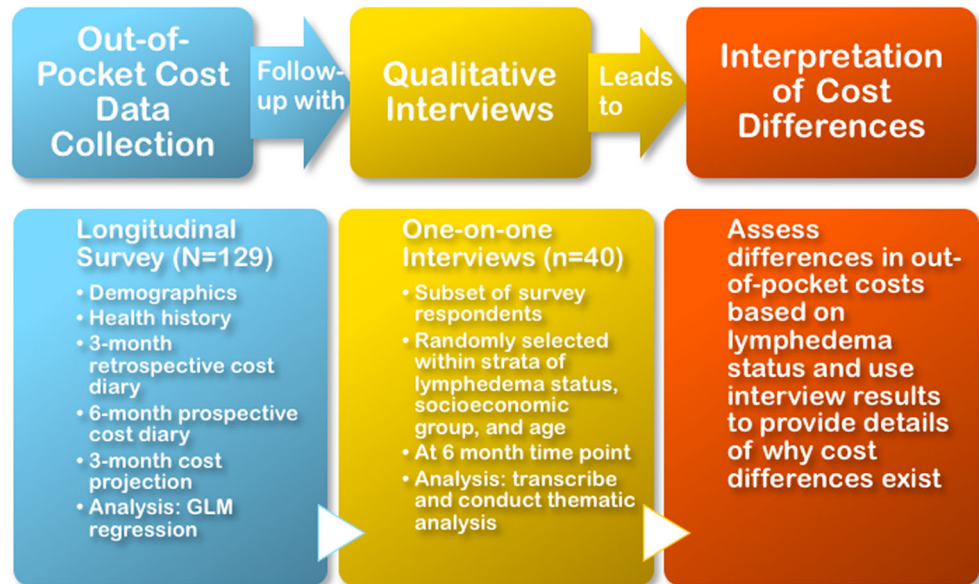
### Measures

**Demographics** Participants self-reported current age, US census-defined race, and socio-economic position. Consumer credit was included because cancer diagnosis has been associated with increased bankruptcy rates [6, 32], which would have an impact on a patient's consumer credit rating for up to 10 years. Self-reported health insurance was classified as public (Medicaid or Medicare), private, or none. Participants reporting both public and private insurance were counted in both categories.

**Costs and productivity losses over 12 months (cost diary)** The data collection instrument developed was based on Goossens' cost diary [33], a validated tool for cost-effectiveness analysis. Participants were instructed to report direct, indirect, and productivity costs related to their overall healthcare including the following: co-payments for outpatient physician visits, physical and occupational therapy visits, complementary and integrative therapy visits, emergency department visits, hospitalizations, labs, X-rays, and tests; wellness resources (e.g., gym memberships); all lymphedema-specific healthcare needs (compression garments, bandages) for lymphedema in any part of the body; medications or other health-related product that a participant identified; and health insurance

**Fig. 1** Conceptual overview of mixed methods approach. This figure shows the flow of the explanatory sequential design of the mixed methods study, which begins with quantitative out-of-pocket costs data collection for all 129 participants, followed by qualitative interviews of 40 randomly selected participants, which were integrated to interpret the cost differences identified between those who had lymphedema and those who did not have lymphedema

## Mixed Methods Analysis of Out-of-Pocket Costs for Long-Term Breast Cancer Survivors with or without Lymphedema: An Explanatory Sequential Design



premiums if paid out-of-pocket (Appendix I in the supplementary material). All out-of-pocket costs were collected in continuous dollar amounts.

Similar to methods used in previously published cost studies of adverse effects of cancer treatment [14], participants reported 12 months of cost data. This was captured through a 3-month retrospective cost diary, then six monthly prospective cost diaries, followed by an estimation of projected costs in the upcoming 3 months. For the 3-month retrospective and 6-month prospective data collection, participants were encouraged to use personal calendars, insurance statements, and receipts to aid in recall. The following 3-month projection was designed to capture any regularly occurring appointment co-pays, such as those for annual check-ups, that did not fall in the earlier time windows that out-of-pocket costs data were being prospectively recorded. Altogether, participants supplied 12 months of cost data and each cost was summed across all the data collection points. For each itemized question, participants with lymphedema designated which costs were related to lymphedema. We considered 12-month costs including and excluding productivity costs to differentiate direct medical and non-medical, and indirect cost domains [34]. Productivity losses for work and for home were calculated based on methods used in a previously published paper that estimated economic burden for US cancer survivors [35] based on self-reported days unable to perform usual activities or hours of help needed to carry out daily activities. For work productivity losses, the adjusted mean number of missed workdays was multiplied by the US Bureau of Labor

Statistics estimates for the median hourly wage (\$16.87) based on a 6-h workday. For household productivity losses, the mean number of hours that someone needed help was multiplied by the daily household productivity rate (\$43.37 per day) based on the consumer price index.

**Subjective ratings of economic burden** At baseline, participants completed a self-administered survey on economic burden using 12 items adapted from the Breast Cancer Finances Survey [36, 37], a validated survey of economically burdensome events among breast cancer survivors that assesses burden related to long-term survival.

**Cancer history and treatments** Participants self-reported completing chemotherapy and/or radiation therapy and/or hormone therapy after cancer surgery, and year of breast cancer diagnosis. Self-report of breast cancer treatment has been validated as over 90% accurate [38]. Whether patients underwent multiple adjuvant treatment modalities is more important than the details of treatment; thus, we modeled the total number of types of treatments.

**Health conditions and lymphedema** Participants self-reported any of 23 comorbidities and previous diagnosis of breast cancer-related lymphedema. To measure upper extremity lymphedema severity, interlimb volume difference measurements between the affected and unaffected arms were taken using perometry (Juzo, Germany), and were adjusted for humidity, barometric pressure, and time of day. Women who

wore compression garments were asked to remove them for at least 1 h prior to perometry assessment.

## Qualitative data collection

As depicted in Fig. 1, the explanatory sequential design [39] of this mixed methods study included 30-min qualitative interviews at the end of monthly prospective out-of-pocket cost data collection. This method entails first collecting quantitative data, and then collecting qualitative data to inform and provide context for quantitative findings. This approach used data from semi-structured interviews to explore what might drive cost differences between breast cancer survivors with and without lymphedema, and capture ways in which long-term economic burden might affect breast cancer survivors that might not be easily quantifiable with a cost diary approach alone. A standardized semi structured interview guide (Appendix II in the supplementary material), developed by the study PI, included questions on economic challenges, supports utilized, lasting impact and resource gaps after participants' breast cancer diagnosis. The study PI and a trained research assistant conducted interviews with a subset of 40 participants who were purposively sampled to ensure equal representation across three sampling categories: lymphedema status, socioeconomic position (high school or less vs. college or more), and age group (over 65 and under 65). The sample included 40 participants to ensure at least 10 participants per each sampling category and the potential to reach saturation of themes. Participants were randomly selected using a random number generator and placed into each sampling category until there were at least ten in each category.

## Data analysis

For quantitative analysis of the entire study sample, baseline statistical differences between demographic characteristics were calculated using Chi-squared test and Fischer's statistic for demographic categories with less than five respondents, and non-parametric Ranksum test for non-normally continuously distributed variables. A generalized linear model (GLM) with the power 0.5 link and negative binomial family was used to account for over dispersion because the cost outcome variables contained several zeros and the variance exceeded the mean distribution, which is common with health care cost data. After the model was fitted, the marginal effects of the total cost for those with and without lymphedema were calculated and graphed, adjusted for each covariate at its mean. Two separate GLM models were estimated: one for the total cost including and one for the total cost excluding productivity costs.

For qualitative analysis of the 40 interviewees, verbatim interview transcripts were inputted into MAXQDA software program for qualitative analysis. First, structural codes based

on interview questions and domains of economic burden among cancer survivors documented in the literature were identified. Additional codes were included after transcripts were reviewed for themes. The research team organized these codes into a codebook, which was used for thematic coding. Each fifth transcript was double coded and discrepancies were discussed and resolved among the research team. Quotes were collected to illustrate key findings and ensure that conclusions were consistent with the data.

## Results

The 129 participants who completed data for the study are described in Table 1. Across the entire sample, the mean age was 63 and the average time since cancer diagnosis was 12 years. Just under half (46.5%) of participants had been diagnosed with lymphedema. There was no statistically significant difference by lymphedema status in mean age, race, education, social status, wealth, credit score, type of insurance, or number of economically burdensome events. A significantly greater percentage of women with lymphedema were in a lower income category ( $p = 0.02$ ) compared to those without lymphedema. Cancer stage at diagnosis, type of adjuvant treatments, and number of comorbidities did not differ by lymphedema status, but those with lymphedema were on average 3 years farther out from diagnosis ( $p = 0.002$ ) and had greater interlimb difference ( $p < 0.001$ ).

Figure 2 compares the average dollar amount difference, with and without productivity losses, for those who did not have lymphedema and those who did, controlling for age, race, socioeconomic position, type of cancer treatments, comorbidities, years since cancer diagnosis, and type of insurance. Excluding productivity losses, participants with lymphedema are estimated to have an average \$2306 in out-of-pocket costs per year compared to \$1090 for those without lymphedema ( $p = 0.006$ ), or 112% higher costs (Fig. 2a). Including productivity losses, participants with lymphedema are estimated to have an average \$3325 in out-of-pocket costs per year compared to \$2792 for those without lymphedema ( $p = 0.55$ ), or 19% higher costs (Fig. 2b). Among those reporting having lymphedema, Fig. 3 shows that nearly 50% of total costs were attributed to lymphedema.

Table 2 contains illustrative quotes that demonstrate the three major themes that emerged from the qualitative interviews comparing breast cancer survivors across lymphedema status. These interviews provide further insight into the burden of higher costs associated with lymphedema.

Theme 1: Economic burden is cumulative and cascades over time; managing an adverse treatment effect presents ongoing challenges. The use of savings to cover medical

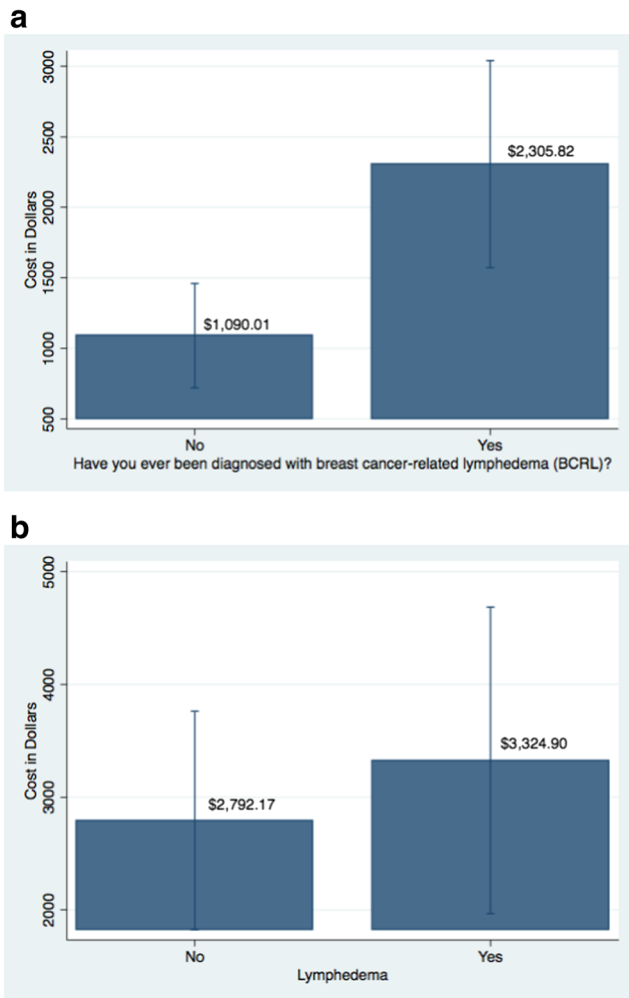
**Table 1** Participant baseline characteristics

<i>N</i> = 129	BCRL yes, <i>n</i> = 60 (46.51%)	BCRL no, <i>n</i> = 69 (54.49%)	<i>p</i> value
<b>Demographics</b>			
Age in years, M (SD)	65 (8)	62 (8)	0.11
Race			0.32
White	35 (57.4)	41 (60.3)	
Black	24 (39.3)	26 (38.2)	
Other	2 (3.3)	0 (0.0)	
Education completed			0.35
High school	17 (27.9)	13 (19.1)	
College	26 (42.6)	29 (42.7)	
Graduate school	17(27.9)	26 (38.2)	
Income			0.02
≤ \$30,000	8 (13.1)	11 (16.2)	
\$30,001–\$70,000	30 (49.2)	18 (26.5)	
> \$70,000	19 (31.2)	35 (51.5)	
Total cash assets			0.60
≤ \$4999	17 (27.9)	16 (23.5)	
\$5000–\$49,999	16 (26.2)	13 (19.1)	
\$50,000–\$499,999	13 (21.3)	19 (27.9)	
≥ 500,000	9 (14.8)	13 (19.1)	
Consumer credit quality ( <i>n</i> = 123)			0.12
Poor/fair	18 (31.6)	12 (18.2)	
Good/very good	22 (38.6)	24 (36.4)	
Excellent	17 (29.8)	30 (45.5)	
Insurance type			
Public	21 (34.4)	19 (27.9)	0.43
Private	49 (80.3)	53 (77.9)	0.74
None	1 (1.6)	2 (2.9)	0.62
Economic burden score [range 0–12] (SD)	3 (3)	2 (4)	0.95
<b>Cancer diagnosis and treatment variables</b>			
Cancer stage at diagnosis			0.09
Stage 0	9 (14.8)	10 (14.7)	
Stage 1	11 (18.0)	22 (32.4)	
Stage 2	11 (16.2)	19 (31.2)	
Stage 3	9 (14.8)	6 (8.8)	
Missing	13 (21.3)	19 (27.9)	
Years since cancer diagnosis (SD)	13 (6)	10 (3)	0.002
Number of adjuvant treatment modalities (SD)	2 (1)	2 (1)	0.13
Radiation	51 (83.6)	53 (77.9)	0.42
Chemotherapy	51 (83.6)	46 (67.7)	0.05
Hormonal therapy	29 (47.5)	34 (50)	0.79
Comorbidities	1 (1)	1 (1)	0.46
Interlimb difference (%)	9.3 (13.4)	−0.8 (6.1)	< 0.001

BCRL breast cancer-related lymphedema

costs and additional loans or debt to cover medical costs was common in all respondent interviews. For some women, covering medical costs compromised their ability to manage basic needs like utility bills. Women with

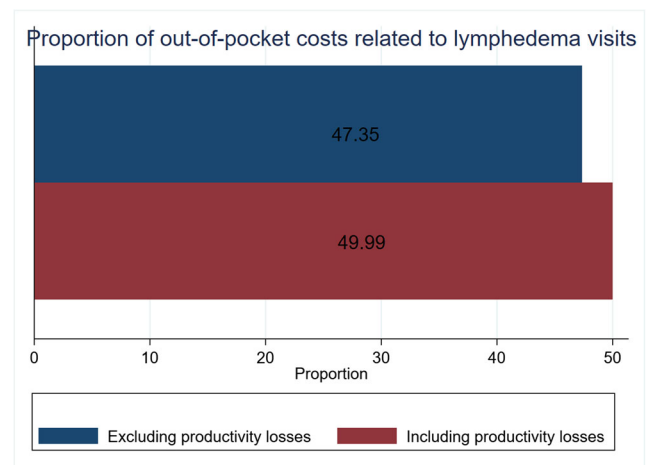
lymphedema were more likely to relay that the upfront costs associated with cancer set off a cascade of financial challenges that continues to affect their current economic situation. Participants described current effects such as



**Fig. 2** Adjusted annual out-of-pocket costs (marginal effects) for breast cancer survivors without or with lymphedema. **a** Mean annual out-of-pocket costs, excluding productivity losses, and 95% confidence intervals. The bar graphs in this figure compare mean annual out-of-pocket costs excluding productivity losses of \$2306 for long-term breast cancer survivors with lymphedema and \$1090 without lymphedema, adjusted for age, race, education, income, cash assets, consumer credit quality, interlimb difference, number of cancer adjuvant treatment modalities, years since diagnosis, number of comorbid conditions, and public or private insurance. **b** Mean annual out-of-pocket costs, including productivity losses, and 95% confidence intervals. The bar graphs in this figure compare mean annual out-of-pocket costs including productivity losses of \$3325 for long-term breast cancer survivors with lymphedema and \$2792 without lymphedema, adjusted for age, race, education, income, cash assets, consumer credit quality, interlimb difference, number of cancer adjuvant treatment modalities, years since diagnosis, number of comorbid conditions, and public or private insurance

decreased ability to help family, support their children's educational endeavors, and retire. Ongoing costs for lymphedema care needs exacerbated economic burden and compromised participants' ability to obtain care for their current lymphedema needs.

Theme 2: Lymphedema care needs are unlikely to be covered by insurance, which contributes to higher long-



**Fig. 3** Proportion of total costs attributed to lymphedema-related needs, among those with lymphedema. The bar graphs show that 47% and 50% of cost, excluding and including productivity losses, respectively, for patients with lymphedema were costs directly relating to lymphedema needs

term costs and compromises a patient's ability to manage lymphedema symptoms. Respondents in both groups reported out-of-pocket health care costs and shifting costs to other parties (including family, employers, social service organizations, and advocacy groups). Participants described the need to use leftovers of patients' medications to cope with their economic burden. Women who did not have lymphedema were more likely to report out-of-pocket costs accrued closer to the period of their cancer treatment for supplemental insurance, co-pays, and treatment, while women with lymphedema reported additional ongoing long-term out-of-pockets costs for lymphedema care in the form of ongoing physical therapy, lymphedema specialists, sleeves, and garments that were not covered by insurance. Even participants with private insurance did not always receive necessary lymphedema-specific care because of the cost burden. Changes in insurance, especially when changes in status led to less lymphedema coverage, further stymied their ability to manage ongoing lymphedema needs.

Theme 3: Productivity losses have long-term impact: breast cancer diagnosis may have influenced work opportunities and long-term earning potential, and breast cancer-related lymphedema may further decrease productivity losses at work. Both sets of participants spoke about long-term productivity losses. In some cases, women missed out on educational opportunities, modified work schedules, experienced job loss, pursued voluntary early retirement, or went back to work sooner than medically recommended. These experiences framed their subsequent health and lifestyles and still affect them currently. Women in both sets recalled needing additional help for duties around the house. Women with lymphedema were

**Table 2** Illustrative quotes from research participants by lymphedema status

Theme	Sub theme	Respondents with lymphedema	Respondents with no lymphedema
Theme 1: Economic burden is cumulative and cascades over time; managing an adverse treatment effect presents ongoing challenges	Use of assets, loans and lasting impact of cost accrual	I had to take my 401 K money and like pay bills, buy medicine because I did not have any medical coverage ... all the moneys that I had saved up that would have sustained me [as a retiree] was gone ... in terms of the money that I would have wanted to contribute [to retirement and children's college funds], you know, I wasn't able to and when I was able to-- I'm 61 so I'll never get caught up with that so, yeah. – age 60; private insurance It still affects our economic situation ... we still feel the effects of the economic problems .... We had the co-pays. We had [lymphedema] therapies, different therapies... Massage. And, of course, you know, the sleeves ... it seems like we can never, ever catch up to have a little bit extra. – age 56; private insurance	So I went for a long time just basically on my savings and family helping me. – age 52; private insurance So and I was able to get a small loan and pay off some bills. So, you know, that helped. – age 69; public & private insurance
	Balancing health costs with utility bill costs	It was just like, just a lot of financial burden so it was stressful where I would have liked to have had the experience while I was convalescing to be like not worried about are my lights gonna get shut off? And sometimes that happened and it was just rough. – age 60; private insurance	... So I was no longer able to work, 'cause I had three surgeries ... And then I had to do chemo and then I did radiation ... I had my lights cut off. I had my water shut off. I had my gas shut off. And I would have to go up to the hospital and get slips to get them cut back on ... And they would cut my lights off for, like, maybe \$100... – age 69; public & private insurance
	Increased costs due to lymphedema--specific health needs	So having to go to physical therapy, it's \$30 each time I go ... So I have had to actually ceased going because I just do not have the money. – age 62; private insurance I just ordered my replacement sleeve on Monday, and I had to give my credit card for \$420 before they would put in that order ... This was one sleeve and glove – age 66; public insurance	
Theme 2: Lymphedema care needs are unlikely to be covered by insurance, which contributes to higher long-term costs, and lack of management of lymphedema symptoms	Insufficiency of Medicaid to cover lymphedema needs	The physical therapy is covered with my Medicare and the secondary insurance, but if I were to get any garments, or new bandages, and everything, I am gonna have to do the out of pocket stuff, and I know that ran into, like, \$95 for the bandages, and then the tape that you buy to wrap the bandages, the Ace, that runs to, like, \$5 a roll. – age 73; public & private insurance I cannot basically afford to buy the compression sleeve... And insurance does not cover it ... I had [private insurance that] did give me one sleeve. Right after that, they changed my health insurance [to Medicare], so it went from getting the sleeve to not getting the sleeve. – age 68; public insurance	I pay for supplemental insurance to cover it, and I am dealing with... Medicare telling me what I can and cannot take ... My supplemental insurance, to help cover the doctors and stuff, is \$227 a month, and then your supplemental to cover your drugs is another \$45 a month. And of course, Medicare's not free. I know everybody acts like it is, but it's not. Last time I looked, it was \$166 bucks every three months. – age 73; public insurance

**Table 2** (continued)

Theme	Sub theme	Respondents with lymphedema	Respondents with no lymphedema
	Out-of-pocket costs that are not covered by insurance	Right after my diagnosis and treatment and surgery, I had lymphedema and severe cording and banding... So I went to a [lymphedema] therapist, who at that time [the insurers] were not paying for that, it wasn't reimbursed, so it was all out of pocket. – age 67; private insurance	The only thing that wasn't covered was ... a shot that was \$100 and for-- I think for someone that's not employed, that would be a difficult fee for them to have to pay – age 55; private insurance When you're first diagnosed, you have to go to a bunch of specialists, and the specialists are \$25 apiece. When you're going three times a week ... it does add up, even with insurance. – age 56; private insurance
Theme 3: Productivity losses have long-term impact: breast cancer diagnosis may have influenced work opportunities and long-term earning potential and breast cancer-related lymphedema may further decrease productivity losses at work	Loss of career opportunities	I actually, I was teaching first grade at the time, which is very physically challenging, and I decided at the end of that school year, in June I retired – age 66; public insurance I lost my job 'cause I got diagnosed with breast cancer so financially it was very difficult ... I was out of work for almost a year ... with the chemo ... I was really sick and then I went back against the doctor's orders 'cause I needed to make money... When I came back to work that's when they expected me to resume all of the duties... full force and ... I got fired... – age 60; private insurance I used to do work with a lady with catering and stuff right and I could not use my arm because it was always in pain with the lymphedema ... It was a setback .... I stopped [working]. – age 63; private insurance	When I went back, [the university] had taken away my financial aid, and consequently I was not able to complete my PhD. That's an enormous hit. Consequently, although I am teaching at the University level ... they will not hire me full time because I do not have the PhD. That would not have happened had I not had cancer... I also had chemo brain at that point... I mean, I still was getting good grades, but it was much harder work, but I also had no money, and we could not afford it, so I quit [the PhD program]. I have regretted that all these years. – age 59; private insurance
	Needing help with daily activities	I just went around my normal household duties, and only thing I didn't do-- I don't think I did any ironing. – age 81; public insurance	During the first year, during treatment and immediately following, one, I was out of work for six months. Two, I needed help with childcare, transportation for children, housekeeping, meal prep. – age 60; private insurance
	Taking time off from work	Well, the surgery, I was-- I think I was out of work for maybe a month. For the lymphedema treatments, I just would go after work. I had to maybe leave early for work and leave early for radiation and that was about six weeks I think – age 63; private insurance I would schedule my chemo on a Friday, so it would give me Saturday and Sunday if I needed it. And, for my radiation, my employer would let me leave like at one o'clock every day... – age 60; public insurance	After I had my [breast cancer surgery] surgery, I wound up back in the hospital with a severe infection... because I did not get, or I did not understand, or I did not hear the proper way to keep it draining. And it backed up, and I wound up in the hospital for another four days with that. – age 73; public insurance I didn't go back to work until part-time in November. So from June to November. And then, full-time, I guess, December or January ... so we had the loss of salary plus additional outlay. – age 60; private insurance



less likely to return to employment after cancer because of their additional physical challenges.

## Discussion

Study results suggest that the economic burden of breast cancer continues long after diagnosis. Women with lymphedema experience a higher burden, with or without indirect cost considerations. In the long term, women report losses to economic opportunity due to their cancer and lymphedema diagnoses. Despite the expansion of cancer-related insurance coverage under the Affordable Care Act, breast cancer survivors, with and without lymphedema, still face significant financial need.

Breast cancer survivors with lymphedema faced up to 112% higher total out-of-pocket costs when excluding productivity losses and up to 19% higher total out-of-pocket costs when including productivity losses, compared to those without lymphedema. The average out-of-pocket costs estimated in this study are lower than the roughly \$11,000 per year previously estimated for US women [13]; however, previous estimates are based on costs closer to the time of treatment, when overall healthcare needs may be higher, and are based on women of working age, who would not have out-of-pockets subsidized by Medicare. These estimates are based on insurance claims, and not patient out-of-pocket costs, which may also contribute to why previous estimates differ from those found in the present study. Even though our costs are lower than previously estimated, the study provides evidence that costs for women with lymphedema remain significantly elevated long after cancer treatment. On average, there is a \$500–\$1215 difference. These economic burdens occur even among those who have health insurance.

Although nearly all of the women in the sample had some form of insurance, changes and challenges with insurance consistently complicated issues for both women who had lymphedema and those who did not. Studies conducted before the 2010 Affordable Care Act reported that financial burden created worry and anger when tools for lymphedema management were not covered by insurance [40]. The present study suggests that these challenges persist even after the Affordable Care Act, which has the potential to expand health insurance coverage for cancer-related care [41] and for cancer survivors [42, 43]. For women cancer survivors with lymphedema, who continued to manage a disease long after completing cancer treatment, these challenges persisted due to ongoing needs for lymphedema care. Women who did not have lymphedema reported insurance challenges related to coverage of cancer treatment and co-pays at the time of treatment but did not report challenges with current care.

Insurance coverage of lymphedema care varied over time and changed based on insurer. Previous findings suggest persistently high costs for cancer survivors who are insured by public insurance [44], which are particularly relevant because coverage for lymphedema treatment varies for public insurance [45]. Medicare covers: medically necessary manual lymph drainage performed by physical or occupational therapists, compression bandaging services, patient education on lymphedema self-care and lymphatic decongestion exercises, and pneumatic compression devices, but not for all compression self-management equipment (such as bandages). Medicaid expands on that coverage, with a few states covering compression garments and bandages. Switching from private to public insurance often posed the most problems when public and private insurance covered lymphedema differently such that patients' needs were no longer met. When patients could not cover their costs, family members or social service organizations were sometimes able to help, but often patients simply went without the care they needed. Other studies have suggested that high out-of-pocket costs will cause patients to use compression garments that no longer apply sufficient pressure to manage lymphedema [46], which participants in our study corroborated. Interview participants reported that lack of coverage for lymphedema-related costs contributed to less lymphedema management and exacerbation of lymphedema.

This study confirmed that higher costs can only be partly attributed to lymphedema, above and beyond the presence of other comorbidities. Interview data suggest that higher costs for women with lymphedema stemmed from cumulatively high economic burden that cascaded over time and prevented women from fully recovering financially. It also highlighted that examining only financial costs due to lymphedema underestimates its full cumulative effect of economic burden on ability to afford other basic needs. Use of savings and retirement to cover lymphedema and healthcare costs can affect women long term and have intergenerational effects. Managing breast cancer-related lymphedema presents ongoing challenges, and adherence can be difficult due to ongoing costs. Having access to additional resources through family, credit, or savings is often leveraged, but may never be recovered, especially for those with ongoing lymphedema management needs.

The findings of this study are especially timely given the recent calls to reduce financial toxicity in US cancer patients through individual-level strategies, as encouraging healthier behaviors [47] and greater financial disclosure [48, 49]. But these strategies put the onus on patients to act in order to reduce cost, rather than pointing to healthcare systems to change to reduce costs. This approach may widen disparities since patients with the greatest resources will be able to afford better health, thus reducing costs is paramount. Some scholars have explored healthcare provider-driven recommendations to reduce economic burden [50]. Other scholars have called for

introducing screening for financial toxicity, as well as a multi-level approach to reducing economic burden [51], and redesigning sick leave policies to better accommodate chronic disease needs [46]. For mitigating lymphedema costs in particular, advocates have supported the Lymphedema Treatment Act, which would amend Title XVIII (Medicare) of the Social Security Act to cover certain lymphedema compression treatment items as durable medical equipment under Medicare. Some states, including California, Louisiana, North Carolina, and Virginia, have issued their own laws, but the Act has not found success at the US Congressional level. A 2016 report found that expanding insurance coverage in one state had a less than 0.1% impact on insurance claims, while lowering costs for lymphedema treatment and lymphedema-related hospitalizations [52]. Our findings reinforce the need for actions at policy, provider, and individual patient levels, especially for those with lymphedema.

This analysis consisted of a small sample from one geographic area, the majority of whom had insurance, which may limit the external validity of the findings, especially given that insurance policies differ regionally. Responses may be different from women living in regions with other insurance offerings. Cost diaries pose a time burden on participants to complete, which may have led to missing entries: monthly text-based, e-mail, and phone messaging was used to remind participants to complete their diaries. Participants were also allowed to send in receipts and medical visit bill summaries in lieu of writing them into the cost diaries themselves. Nonetheless, the data that were collected comprehensively covered cost domains in real time, with over a 90% response rate in each month of data collected. Cost data collection was not prospective throughout, and the 3-month retrospective data may have been biased due to recall; to minimize the potential for bias, participants were asked to use supporting documents to aid in recall. The final 3 months were based on projected costs due to regular ongoing medical visits or needs, leading to underestimated costs because unexpected healthcare needs would not be included. Cost diaries may not comprehensively capture the various domains of direct, indirect, psychosocial, and time costs and only measure costs over the period of observation; thus, we used qualitative data to supplement our understanding of how and when various types of costs were incurred during the course of survivorship. These challenges mean that the cost estimates are conservative, and that actual out-of-pocket costs are likely higher than reported here. There may have been other clinical factors to consider that may influence cost, like cancer severity [53] as the initial economic shock that set patients on different financial trajectories. Data on stage at cancer diagnosis were excluded from the regression analysis due to a high percentage of missing data (16%), although available data suggest no difference in stage of diagnosis by lymphedema status. As a voluntary research study, those experiencing the greatest

economic or health challenges may not have had time to enroll and participate, meaning that our results may underestimate economic burden. Results may not be generalizable to those with higher cancer stage, older age, or other tumor sites other than the breast, but breast cancer is among the most economically burdensome cancers.

Breast cancer survivors with lymphedema face higher costs than those who do not have lymphedema, even many years after cancer diagnosis. Although women with and without lymphedema experience a similar number of economically burdensome events and comorbidities, high out-of-pocket costs for women with breast cancer-related lymphedema lead to a cascade of other economic challenges that persist long after cancer treatment. Future work should explore patient-driven recommendations to reduce economic burden after cancer.

**Funding information** This work was supported by the National Center for Research Resources and the National Center for Advancing Translational Sciences (NCATS) of the National Institutes of Health through grant 1UL1TR001079 (statistical consultation); the National Cancer Institute grant K01CA184288 (Lorraine T. Dean); the National Institute of Mental Health grant R25MH083620 (Lorraine T. Dean, Yusuf Ransome); the Sidney Kimmel Cancer Center grant P30CA006973 (Lorraine T. Dean, Kala Visvanathan, Yuehan Zhang); Johns Hopkins University Center for AIDS Research grant P30AI094189 (Lorraine T. Dean); the National Institute on Drug Abuse grant T32DA031099 (Shadiya L. Moss), National Institutes of Mental Health K01MH111374 (Yusuf Ransome); the National Institute on Aging grant K01AG04176 (Lauren Hersch Nicholas); and the National Cancer Institute grants R01CA106851 and 1U54CA155850-01 (Kathryn H. Schmitz).

## Compliance with ethical standards

**Ethical approval** All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

**Informed consent** Informed consent was obtained from all individual participants included in the study.

**Conflict of interest** The authors declare that they have no conflict of interest.

**Open Access** This article is distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 International License (<http://creativecommons.org/licenses/by-nc/4.0/>), which permits any noncommercial use, distribution, and reproduction in any medium, provided you give appropriate credit to the original author(s) and the source, provide a link to the Creative Commons license, and indicate if changes were made.

## References

1. Altice CK, Banegas MP, Tucker-Seeley RD, Yabroff KR (2017) Financial hardships experienced by cancer survivors: a systematic review. *J Natl Cancer Inst* 109(2):djw205

2. Zafar SY, Peppercom JM, Schrag D, Taylor DH, Goetzinger AM, Zhong X, Abernethy AP (2013) The financial toxicity of cancer treatment: a pilot study assessing out-of-pocket expenses and the insured cancer patient's experience. *Oncologist* 18(4):381–390. <https://doi.org/10.1634/theoncologist.2012-0279>
3. Bernard DS, Farr SL, Fang Z (2011) National estimates of out-of-pocket health care expenditure burdens among nonelderly adults with cancer: 2001 to 2008. *J Clin Oncol* 29(20):2821–2826
4. Guy GP, Yabroff KR, Ekwueme DU, Virgo KS, Han X, Banegas MP, Soni A, Zheng Z, Chawla N, Geiger AM (2015) Healthcare expenditure burden among non-elderly cancer survivors, 2008–2012. *Am J Prev Med* 49(6):S489–S497
5. Arozullah AM, Calhoun EA, Wolf M, Finley D, Fitzner KA, Heckinger EA, Gorby NS, Schumock GT, Bennett CL (2004) The financial burden of cancer: estimates from a study of insured women with breast cancer. *The Journal of Supportive Oncology* 2(3):271–278
6. Banegas MP, Guy GP, de Moor JS, Ekwueme DU, Virgo KS, Kent EE, Nutt S, Zheng Z, Rechis R, Yabroff KR (2016) For working-age cancer survivors, medical debt and bankruptcy create financial hardships. *Health Aff* 35(1):54–61
7. Bennett CL, Calhoun EA (2007) Evaluating the total costs of chemotherapy-induced febrile neutropenia: results from a pilot study with community oncology cancer patients. *Oncologist* 12(4):478–483
8. Bilir SP, Ma Q, Zhao Z, Wehler E, Munakata J, Barber B (2016) Economic burden of toxicities associated with treating metastatic melanoma in the United States. *Am Health Drug Benefits* 9(4):203–213
9. Irwin DE, Masaquel A, Johnston S, Barnett B (2016) Adverse event-related costs for systemic metastatic breast cancer treatment among female Medicaid beneficiaries. *J Med Econ* 19(11):1027–1033
10. Hansen RN, Ramsey SD, Lalla D, Masaquel A, Kamath T, Brammer M, Hurvitz SA, Sullivan SD (2014) Identification and cost of adverse events in metastatic breast cancer in taxane and capecitabine based regimens. *Springerplus* 3(1):259
11. Hurvitz S, Guerin A, Brammer M, Guardino E, Zhou Z-Y, Viau DL, Wu EQ, Lalla D (2014) Investigation of adverse-event-related costs for patients with metastatic breast cancer in a real-world setting. *Oncologist* 19(9):901–908
12. Rashid N, Koh HA, Baca HC, Lin KJ, Malecha SE, Masaquel A (2016) Economic burden related to chemotherapy-related adverse events in patients with metastatic breast cancer in an integrated health care system. *Breast Cancer (Dove Med Press)* 8:173
13. Shih Y-CT XY, Cormier JN, Giordano S, Ridner SH, Buchholz TA, Perkins GH, Elting LS (2009) Incidence, treatment costs, and complications of lymphedema after breast cancer among women of working age: a 2-year follow-up study. *J Clin Oncol* 27(12):2007–2014. <https://doi.org/10.1200/JCO.2008.18.3517>
14. Schmitz KH, DiSipio T, Gordon LG, Hayes SC (2015) Adverse breast cancer treatment effects: the economic case for making rehabilitative programs standard of care. *Support Care Cancer* 23(6):1807–1817
15. Davidoff AJ, Erten M, Shaffer T, Shoemaker JS, Zuckerman IH, Pandya N, Tai MH, Ke X, Stuart B (2013) Out-of-pocket health care expenditure burden for Medicare beneficiaries with cancer. *Cancer* 119(6):1257–1265
16. Norman SA, Localio AR, Potashnik SL, Torpey HAS, Kallan MJ, Weber AL, Miller LT, DeMichele A, Solin LJ (2009) Lymphedema in breast cancer survivors: incidence, degree, time course, treatment, and symptoms. *J Clin Oncol* 27(3):390–397
17. Paskett ED (2015) Symptoms: lymphedema. In: Ganz PA (ed) *Improving outcomes for breast cancer survivors*. Springer International Publishing AG, Cham, pp 101–113
18. American Cancer Society (2016) *Cancer treatment and survivorship facts and figures 2016–2017*. American Cancer Society, Atlanta
19. Mortimer PS (1998) The pathophysiology of lymphedema. *Cancer* 83:2798–2802
20. Rönkä RH, Pamilo MS, von Smitten KA, Leidenius MH (2004) Breast lymphedema after breast conserving treatment. *Acta Oncol* 43(6):551–557
21. Johansson K, Klemas P, Weibull A, Mattsson S (2014) A home-based weight lifting program for patients with arm lymphedema following breast cancer treatment: a pilot and feasibility study. *Lymphology* 47(2):51–64
22. Haida A, Kuehnb T, Konstantiniukc P, Köberle-Wührera R, Knauera M, Kreienbergd R, Zimmermanna G (2002) Shoulder-arm morbidity following axillary dissection and sentinel node biopsy for breast cancer. *Eur J Surg Oncol* 28:705–710
23. Erickson VS, Pearson ML, Ganz PA, Adams J, Kahn KL (2001) Arm edema in breast cancer patients. *J Natl Cancer Inst* 93(2):96–111
24. Ahmed RL, Prizment A, Lazovich D, Schmitz KH, Folsom AR (2008) Lymphedema and quality of life in breast cancer survivors: the Iowa women's health study. *J Clin Oncol Off J Am Soc Clin Oncol* 26(35):5689–5696. <https://doi.org/10.1200/JCO.2008.16.4731>
25. Lam R, Wallace A, Burbidge B, Franks P, Moffatt C (2006) Experiences of patients with lymphoedema. *J Lymphoedema* 1(1):16–21
26. Fantoni SQ, Peugniez C, Duhamel A, Skrzypczak J, Frimat P, Leroyer A (2010) Factors related to return to work by women with breast cancer in northern France. *J Occup Rehabil* 20(1):49–58
27. Mehnert A (2011) Employment and work-related issues in cancer survivors. *Crit Rev Oncol Hematol* 77(2):109–130
28. Albright HW, Moreno M, Feeley TW, Walters R, Samuels M, Pereira A, Burke TW (2011) The implications of the 2010 patient protection and affordable care act and the health care and education reconciliation act on cancer care delivery. *Cancer* 117(8):1564–1574
29. Schmitz KH, Ahmed RL, Troxel AB, Cheville A, Lewis-Grant L, Smith R, Bryan CJ, Williams-Smith CT, Chittams J (2010) Weight lifting for women at risk for breast cancer-related lymphedema: a randomized trial. *Jama* 304(24):2699–2705
30. Schmitz KH, Troxel AB, Cheville A, Grant LL, Bryan CJ, Gross CR, Lytle LA, Ahmed RL (2009) Physical activity and lymphedema (the PAL trial): assessing the safety of progressive strength training in breast cancer survivors. *Contemporary Clinical Trials* 30(3):233–245
31. Patterson RE, Colditz GA, Hu FB, Schmitz KH, Ahima RS, Brownson RC, Carson KR, Chavarro JE, Chodosh LA, Gehlert S (2013) The 2011–2016 transdisciplinary research on energetics and cancer (TREC) initiative: rationale and design. *Cancer Causes Control* 24(4):695–704
32. Ramsey S, Blough D, Kirchhoff A, Kreizenbeck K, Fedorenko C, Snell K, Newcomb P, Hollingworth W, Overstreet K (2013) Washington state cancer patients found to be at greater risk for bankruptcy than people without a cancer diagnosis. *Health Aff* 32(6):1143–1152
33. Goossens M, Ruten-van Molken M, Vlaeyen J, van der Linden S (2000) The cost diary: a method to measure direct and indirect costs in cost-effectiveness research. *J Clin Epidemiol* 53(7):688–695
34. Brown ML, Yabroff KR (2006) Chapter 12: economic impact of cancer in the United States. In: Fraumeni J, Schottenfeld D (eds) *Cancer epidemiology and prevention*. Oxford University Press, New York, p 202
35. Guy GP Jr, Yabroff KR, Ekwueme DU, Rim SH, Li R, Richardson LC (2017) Economic burden of chronic conditions among survivors of cancer in the United States. *J Clin Oncol* 35(18):2053–2206

36. Gordon L, Scuffham P, Hayes S, Newman B (2007) Exploring the economic impact of breast cancers during the 18 months following diagnosis. *Psychooncology* 16:1130–1139
37. Given BA, Given CW, Stommel M (1994) Family and out of pocket costs for women with breast cancer. *Cancer Pract* 2(3):187–193
38. Oberst K, Bradley CJ, Schenk M (2008) Breast and prostate cancer patient's reliability of treatment reporting. *J Registry Manag* 36(1): 12–15
39. Creswell JW, Plano Clark VL, Gutmann ML, Hanson WE (2003) Handbook of mixed methods in social and behavioral research. In: Tashakkori A, Teddlie C (eds) *Advanced mixed methods research designs*, vol 209. SAGE Publications, Thousand Oaks, p 240
40. Fu MR, Ridner SH, Hu SH, Stewart BR, Cormier JN, Armer JM (2013) Psychosocial impact of lymphedema: a systematic review of literature from 2004 to 2011. *Psycho-Oncology* 22(7):1466–1484
41. Moy B, Polite BN, Halpern MT, Stranne SK, Winer EP, Wollins DS, Newman LA (2011) American Society of Clinical Oncology policy statement: opportunities in the patient protection and affordable care act to reduce cancer care disparities. *J Clin Oncol* 29(28): 3816–3824
42. Nikpay SS, Tebbs MG, Castellanos EH (2018) Patient protection and affordable care act Medicaid expansion and gains in health insurance coverage and access among cancer survivors. *Cancer* 124(12):2645–2652. <https://doi.org/10.1002/cncr.31288>
43. Parsons HM, Schmidt S, Tenner LL, Bang H, Keegan TH (2016) Early impact of the patient protection and affordable care act on insurance among young adults with cancer: analysis of the dependent insurance provision. *Cancer* 122(11):1766–1773. <https://doi.org/10.1002/cncr.29982>
44. Narang AK, Nicholas LH (2016) Out-of-pocket spending and financial burden among Medicare beneficiaries with cancer. *JAMA Oncology* 3(6):757–765
45. Dupas P (2011) Health behavior in developing countries. *Annu Rev Econ* 3(1):425–449
46. Boyages J, Xu Y, Kalfa S, Koelmeyer L, Parkinson B, Mackie H, Viveros H, Gollan P, Taksa L (2017) Financial cost of lymphedema borne by women with breast cancer. *Psycho-Oncology* 26(6):849–855
47. Kaul S, Avila JC, Jupiter D, Rodriguez AM, Kirchhoff AC, Kuo Y-F (2017) Modifiable health-related factors (smoking, physical activity and body mass index) and health care use and costs among adult cancer survivors. *J Cancer Res Clin Oncol* 143(12):2469–2480
48. Zafar SY (2016) Financial toxicity of cancer care: it's time to intervene. *J Natl Cancer Inst* 108(5):djv370. <https://doi.org/10.1093/jnci/djv370>
49. Zafar SY, Abernethy AP (2013) Financial toxicity, part I: a new name for a growing problem. *Oncology (Williston Park)* 27(2): 80–81 149
50. Zafar SY, Newcomer LN, Jusfin McCarthy J, Nasso SF, Saltz LB (2017) How should we intervene on the financial toxicity of cancer care? One shot, four perspectives. *Am Soc Clin Oncol Educ Book* 37:35–39
51. Khera N, Holland JC, Griffin JM (2017) Setting the stage for universal financial distress screening in routine cancer care. *Cancer* 123(21):4092–4096
52. Weiss R (2016) Cost of a lymphedema treatment mandate-10 years of experience in the Commonwealth of Virginia. *Heal Econ Rev* 6(1):42
53. Schwartz KL, Simon MS, Bylsma LC, Ruterbusch JJ, Beebe-Dimmer JL, Schultz NM, Flanders SC, Barlev A, Fryzek JP, Quek RG (2018) Clinical and economic burden associated with stage III to IV triple-negative breast cancer: a SEER-Medicare historical cohort study in elderly women in the United States. *Cancer* 124(10):2104–2114