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A retrospective cost-effectiveness analysis of different cognitive-behavioral therapy for insomnia intervention delivery approaches in adult cancer survivors

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Abstract

Background: Cognitive-behavioral therapy for insomnia (CBT-I) is considered the gold standard treatment for insomnia. Prior trials have delivered CBT-I across a range of treatment sessions. Understanding the economics of varying treatment approaches is essential for future implementation considerations.

Methods: We conducted a retrospective cost-effectiveness analysis from the provider's perspective, comparing the implementation of a three-session CBT-I program for cancer survivors (CBT-I-CS) versus a stepped care treatment approach consisting of an initial single sleep education session followed by CBT-I-CS if elevated insomnia symptoms persisted. The effectiveness measure used was the percentage of participants whose insomnia had remitted by the end of each program.

Results: Stepped care delivery was more effective than CBT-I-CS alone, resulting in 35.4% more remitted patients by the end of the overall program. For a $480 willingness to pay threshold per percentage of remitted patients, stepped care CBT-I-CS reached a 98% probability of being cost-effective, while CBT-I-CS alone had only a 2% probability. Larger group sessions in the first step of a stepped care delivery model resulted in more favorable cost-effectiveness.

Conclusions: A stepped care delivery model may be a more cost-effective approach if it can be implemented efficiently. These findings inform policies aimed at improving cancer survivors' access to much-needed insomnia treatment in settings where financial resources for CBT-I may be limited, and be an important barrier to treatment dissemination.

Clinical Trial Registration: These analyses were not registered.

KEYWORDS
cancer survivors, cognitive-behavioral therapy for insomnia implementation, cost-effectiveness, insomnia, oncology setting, psycho-oncology, stepped care

Eric S. Zhou and Wen You are Co-senior authors.

1 | INTRODUCTION

The number of cancer survivors in the U.S. is projected to exceed 22 million by 2030.[1] They are at higher risk for physiological and psychological issues compared to individuals without cancer.[2,3] Insomnia, defined as the difficulty in initiating or maintaining sleep resulting in daytime sequelae, is one of the most distressing and common consequences of their treatments,[4] affecting up to 30% of survivors.[5,6]

Cognitive behavior therapy for insomnia (CBT-I) is considered the first-line treatment for insomnia disorder.[7] CBT-I is a multicomponent intervention including sleep restriction, stimulus control, sleep hygiene, and cognitive restructuring of maladaptive sleep-related beliefs. The standard treatment protocol typically involves six intervention sessions over 4–6 months. There is consistent data demonstrating the efficacy of CBT-I in cancer survivors.[8] One of the critical barriers to the widespread implementation of this evidence-based treatment is the costs associated treatment, both from the provider's and patient's perspective.[9] In particular, the financial costs associated with CBT-I in the oncology setting are poorly understood.[10] In the limited work done, Savard et al.[11] compared face-to-face CBT-I versus video-based CBT-I for 161 women with breast cancer. From the patient's perspective, face-to-face therapy cost 5.5 times more than self-administered video-based CBT-I. This considered direct costs, such as insurance premiums and transportation to the doctor's office, as well as indirect costs, such as lower quality-of-life.

Notably, the cost-effectiveness of insomnia treatments from the healthcare provider's perspective has not been well-studied. This perspective relates to the total costs associated with treatment provision, regardless of who the payer is. This information is critical for decision-makers at the administrative level, and is essential for assessing the feasibility of implementing evidence-based insomnia care within the cancer setting.[10] To address this key gap in the literature, we sought to retrospectively quantify the comparative cost-effectiveness, from the provider's perspective, of a brief CBT-I group intervention versus a stepped care treatment approach.

2 | METHODS

2.1 | Previous intervention trials

2.1.1 | Trial I: CBT-I for cancer survivors

Trial I tested CBT-I for Cancer Survivors (CBT-I-CS), an abbreviated version of standard CBT-I that was also tailored for cancer survivors.[12] Rather than the standard six sessions of CBT-I, CBTI-CS delivered an abbreviated, in-person group program over three biweekly 1-h sessions. The key components of CBT-I were presented in session and supplemented with a self-guided instructional workbook. Sessions were conducted by a single doctoral-level program facilitator. A total of 79 adult cancer survivors were recruited in 2014–2015. After eligibility screening, 38 survivors enrolled and were treated in convenience groups of varying sizes (8, 16, and 14). A total of 29 participants completed all three sessions, with seven dropping out after session 1 and two dropping out after session 2. To ensure comparability between the two studies, the cost-effectiveness analysis (CEA) was based on data from the 23 completers with elevated baseline **insomnia** (see "Effectiveness measures" section). Table 1 details the characteristics of the completers. Participants were asked to complete self-report measures at pre- and post-intervention.

2.1.2 | Trial II: Stepped care trial

Trial II sought to further reduce patient burden by delivering treatment in a stepped care approach.[13] Stepped care principles have been widely applied with psychological treatments and may be a useful service delivery model for insomnia patients.[14] In Trial II, the first step intervention was the Single Session Sleep Education Intervention (SSSEI), a single 1-h group education session, focused on teaching good sleep hygiene practices and helping participants set achievable sleep goals. After receiving the SSSEI, participants' insomnia symptoms were monitored for 3 months, and those whose insomnia symptoms remained clinically elevated were offered the CBT-I-CS program as the second step intervention in the trial. A total of 163 cancer survivors were screened for study inclusion in 2017–2018, with 51 ultimately enrolling. A total of 24 sleep education sessions, with convenience group sizes ranging from 1 to 4 participants, were delivered. A total of 14 cancer survivors whose insomnia persisted after completing the SSSEI received CBT-I-CS, with all 14 completing all three CBT-I-CS sessions.

2.2 | Measures

2.2.1 | Effectiveness measures

The Insomnia Severity Index (ISI) is a 7-item self-report instrument that measures insomnia symptoms.[15] ISI scores range from 0 to 28,

TABLE 1 Participant characteristics.

Characteristics	CBT-I-CS Trial I	Stepped care Trial II
Age (years)	53.4	54.4
Gender (number)		
Female	20 (87.0)	49
Male	3 (13.0)	6
Years since diagnosis	5.38	10.32
Diagnosis		
Breast cancer, n (%)	15 (65.2)	35 (63.6)
Other cancer types	8 (34.8)	20 (36.4)
Number of patients	23	51

with a score <12 indicative of remitted insomnia in clinical samples.[16] In Trial I, ISI scores were collected at both the first and last intervention sessions. In Trial II (stepped-care), baseline and follow-up ISI scores were collected for each step of the intervention. Specifically, in the first step, the ISI was collected prior to the SSSEI, and again 4 and 8-week post, and in the second step the ISI was collected prior to the CBTI-CS intervention and again 4-week post. To ensure comparability, we defined effectiveness as the percentage of participants whose insomnia had remitted (ISI score <12) at the post-intervention timepoint in the CEA for both trials.[16] Further, in this analysis we restricted the Trial I sample to survivors with baseline ISI score ≥12, as this was an eligibility requirement for Trial II. In our case, the benefits (percentage of remitted participants) largely accrued post-intervention; therefore, it was reasonable to use the trial-based multiple short-term time horizons (1–2 months) in our CEA.

2.2.2 | Cost measures (intervention delivery costs)

The costs of implementing each delivery model were estimated using the program construction details provided by the primary investigators (ESZ and CJR). The intervention program costs were derived from the resources required for running the group sessions, namely: time required for session preparation, interventionist time, venue cost, etc. The cost of each session was calculated based on the number of group sessions and the labor cost for the CBT-I protocol delivery personnel (determined from the national average hourly wage of the appropriate occupation level). A research assistant (≥bachelor's level) was needed for the pre-session preparations (1 h per group session), participant instruction (10 min per participant), and preparing workbooks for participants (15 min per participant in session 1). Before the intervention was delivered, 20 h of training was required to prepare a research assistant with no background in sleep interventions to ensure implementation fidelity in conducting program delivery tasks, such as calculating sleep metrics from participant sleep diaries. The trainer was a PhD-level individual with prior experience in sleep interventions. The wage rates for the occupation levels were taken from publicly available data from the U. S. Bureau of Labor Statistics (BLS) and calculated in U.S. dollars for base year 2019 using the BLS inflation calculator. Session costs were computed from the unit price of each resource. Table 2 reports the unit price of resources in 2019 and the total resource cost for each intervention.

2.3 | Trial participants

In both trials, participants were in their early fifties on average, majority female, and primarily breast cancer survivors. Trial II participants were generally farther off-treatment: they were on average 10 years post-diagnosis cancer diagnosis, as compared to 6 years for those in Trial I. Table 1 reports the characteristics of participants in the two trials. In this study, we used published data for both trials that was already IRB approved for publication.

2.4 | Statistical methods

2.4.1 | Incremental cost-effectiveness ratio and cost-effectiveness plane

We evaluated the comparative cost-effectiveness of the two trials using the incremental cost-effectiveness ratio (ICER), the point estimate of the additional cost for an incremental improvement in outcome when comparing the mean costs and outcomes of the two interventions (I1 and I2). The ICER is defined as:

$$\text{ICER} = \frac{\text{Cost}_{I2} - \text{Cost}_{I1}}{\text{Benefit}_{I2} - \text{Benefit}_{I1}}$$

Specifically, the ICER here refers to the difference of trial costs between the two trials divided by the difference in the trials' effects (i.e., percentage of participants whose insomnia remitted). Negative ratios can be interpreted as reduced costs and positive effects but can also indicate higher costs coupled with worse outcomes. Due to imperfect information, both the costs and effects of trials are associated with some degrees of uncertainty. To address these uncertainties in the analysis, stochastic bootstrap resampling (i.e., random sampling with replacement from the trial participants) was used to obtain the joint distribution of incremental costs and effects of trials, and the results are displayed in the cost-effectiveness plane.

Cost-effectiveness planes, usually used to facilitate choices, are graphed with incremental costs (cost difference between trials) on the *y*-axis and incremental effects (difference between outcomes of trials) on the *x*-axis. Depending on the location of the cost/effect pair (i.e., ICER) in the plane, the relative cost-effectiveness of the two trials can be compared. Willingness-to-pay (WTP) is the maximum monetary amount that could be spent to achieve the plausible outcome. Given various WTP thresholds, the cost-effectiveness acceptability curve (CEAC) displays the probability of one trial being more cost-effective than the other.[17] This probability is calculated from the incremental cost-effectiveness plane with reference to the defined WTP threshold; it is the portion of ICER points below the WTP value.

2.4.2 | Sensitivity analysis

Both trials were designed to be delivered as group-based interventions. Most trial costs were related to group implementation, including labor costs and space rental (Table 2). Group session delivery costs can substantially vary for the same program: larger group sizes are associated with fewer sessions and lower overall delivery costs. The group sizes in the two trials were based on feasible

TABLE 2 Intervention costs for the two trials.

Description	Unit price in 2019	Trial I CBT-I-CS intervention	Trial II (stepped care): Single sleep education intervention (SSSEI) session	Trial II (stepped care): CBT-I-CS intervention
Clinic space[a]	$110/h	$1882.26	$5280.00	$660.00
Clinician[b]	$28.17/h	$265.41	$676.08	$84.51
Assistant for sleep diary calculation, instruct patient, room prep, and other tasks	$19.73/h	$1087.40	$882.37	$359.25
Workbook preparation (color printing, regular binding, etc.)	$0.045/page	$18.40	$45.90	$12.60
Patient parking	$7/h	$721.74	$714.00	$588.00
Training for sleep diary calculations				
Trainer (PhD level)[c]	$38.87/h	$789.60	-	$777.40
Assistant (no requirements)[d]	$19.73/h	$381.40	-	$394.60
Total costs	-	$5146.21	$7598.35	$2876.36

Note: The maximum capacity of the venue for group sessions was 25 people.

[a]The maximum capacity of the venue for group sessions was 25 people.

[b]The minimum required education was master's level. We considered the wage rate for a general therapist.

[c]Wage rate for a PhD-level therapist was considered.

[d]We used a staff-level assistant with no minimum education required (office support staff occupation).

recruitment logistics; therefore, they represent realistic variations as expected in real world delivery settings.

We conducted sensitivity analysis to examine the impacts of group size on the comparative cost-effectiveness between the two trials. Since the Trial II offered very small-size group sessions in the SSSEI to accommodate recruitment logistics, we examined how larger group sizes in the SSSEI would affect cost-effectiveness, while keeping constant the size of CBT-I-CS groups delivered either in Trial 1 or Trial II.

3 | RESULTS

3.1 | Effectiveness

The effectiveness of each program was measured as the percentage of participants whose insomnia had remitted at the post-intervention timepoint. In Trial I, 39.1% of participants reported remitted insomnia. In Trial II, 50.9% saw their insomnia remitted after completing the SSSEI session, and an additional 23.5% had remitted insomnia after receiving CBT-I-CS in the second step of Trial II. In total, 74.5% of participants had remitted insomnia by the end of the Trial II. Therefore, there was a larger proportion of remitted participants in Trial II (74.5%) than in Trial I (39.1%; $p < 0.001$).

3.2 | Intervention delivery costs

Table 2 provides a breakdown of implementation costs. The major cost of intervention delivery was related to labor. Overall, Trial I cost $5146.21 to implement, while Trial II cost $7598.35 for the SSSEI intervention in step 1 and $2876.36, for CBT-I-CS sessions in the second step. The SSSEI session in Trial II imposed the highest expense because of the large number of groups. To accommodate recruitment logistics, treatment group session sizes ranged from 1 to 4 participants. With a total of 51 recruited individuals for the stepped care trial, the SSSEI session of Trial II was held 24 times in total, with an average of 2.1 participants/session.

3.3 | Comparative cost-effectiveness

The cost per percent of participants whose insomnia remitted was estimated at $131.51 for Trial I and $140.58 for Trial II. Furthermore, costs per completed participant (regardless of whether their insomnia improved) were estimated at $223.75 and $205.39 for Trial I and Trial II, respectively.

Table 3 details CEA results, including costs, effects, and ICERs, for both trials. Relative to Trial I the stepped care approach used in Trial II was more effective, resulting in 35.4% more remitted participants, while also costing $5301.22 more in total implementation costs. ICER analyses showed that for a single percent increase in the proportion of remitted participants, delivery of CBT-I-CS as the second step in Trial II cost an additional $172.99 (95% CI: $147.81, $198.19) as compared to CBT-I-CS in Trial I.[18] Figure 1A presents the cost-effectiveness plane of the two trials; almost all data points were located in the north-east quadrant, indicating that the stepped care approach of Trial II generated better health outcomes at higher costs than Trial I. Figure 1B displays the CEAC, showing the cost-effective probability for each

of the two trials under various WTP thresholds. Under low WTP (e.g., less than about $100 per percentage of remitted participants), CBT-I-CS delivered in Trial I had much higher probability of being cost-effective than the stepped care approach of Trial II, but its acceptability declined rapidly as the WTP per percentage of remitted participants increased. As Figure 1B shows, when the WTP per percentage of remitted participants reached $150/% remitted, the two trials reached the same probability of cost-effectiveness (about 50%). However, the acceptability of stepped care increased rapidly as the WTP threshold increased beyond this crossing point, and when the WTP was $480/% remitted, the probability of Trial II being cost-effective reached 98% while the probability for Trial I dropped to near 0%.

3.4 | Sensitivity analysis

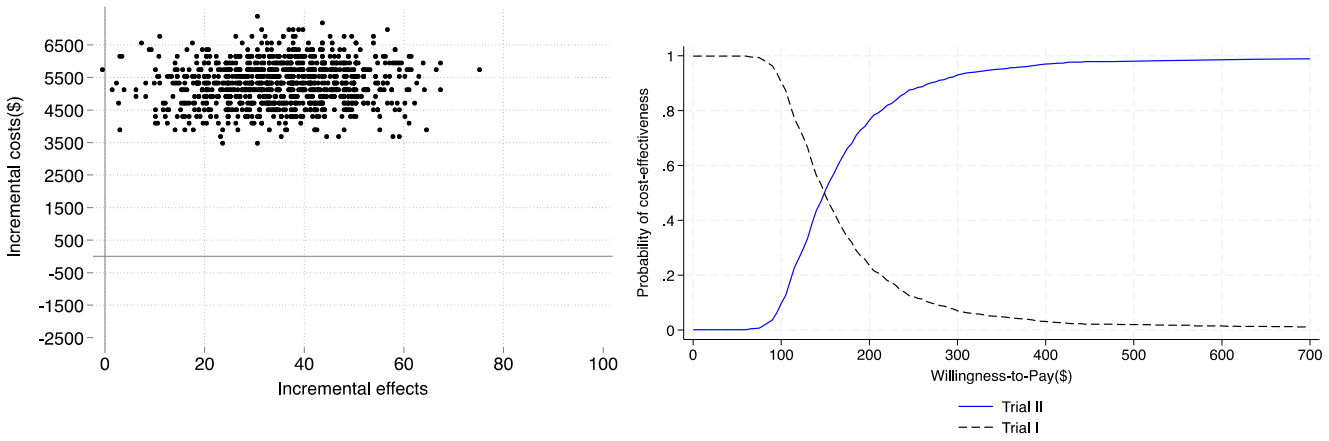
The costliest components in both trials were the resources required to deliver the groups, including clinic space and labor time (Table 2). The main reason the stepped care delivery in Trial II was costlier than Trial I in the cost-effectiveness sense (i.e., requires higher WTP to be acceptable) was due to the small SSSEI group sizes in Trial II. In the sensitivity analysis, we varied the number of patients in the SSSEI sessions from 1 to 25.[19] Figure 2 shows the corresponding ICER for varying group sizes of the SSEI sessions while holding constant those of CBT-I-CS sessions in both trials. As expected, larger group sizes resulted in decreases in the number of group sessions held, cutting down the intervention costs drastically. As Figure 2 reveals, the ICER becomes negative (favoring the stepped care program) with a minimum of 13 participants per session for the SSSEI session of the stepped care delivery.

The ICER plane and CEAC corresponding to the scenario with ≥13 participants per SSSEI session (with 51 participants in Trial II which results in a total of 4 repeated SSSEI sessions to be held) in Trial II are shown in Figure 3A,B, respectively. The points in the ICER plane (Figure 3A) are scattered in both the northeast and southeast quadrants. The points in the southeast quadrant indicate cases where the stepped care approach in Trial II is more effective at a lower cost, as compared to Trial I. This can also be seen in Figure 3B: at zero WTP, there is a 57% chance for Trial II to be more cost-effective than delivering CBT-I-CS as in Trial I. In other words, the stepped care delivery in Trial II with ≥13 participants per the SSEI session is first-order stochastically dominant over Trial I, meaning that regardless of the maximum WTP level, with a minimum of 13 participants per the SSSEI session, decision makers will always prefer the stepped care approach.

TABLE 3 cost-effectiveness analysis: Bootstrapped results.

Analysis	ΔC[a]	ΔE[b]	ICER ($/% remitted)[c]
Base case scenario	$5301.22 [95% CI: $5260.43, $5341.10]	35.4	172.99 [95% CI: 147.81, 198.19]
SA[d] (larger group size in the SSSEI of the trial II[e])	−$56.79 [95% CI: −97.57, −16.00]	35.4	−3.84 [95% CI: −6.32, −1.32]

[a]ΔC: cost differences between Trial II (i.e., the SSSEI in step 1 and CBT-I-CS intervention in step 2) and Trial I (i.e, CBT-I-CS intervention).
[b]ΔE: difference in effects between Trial II and Trial I. The effect is defined as the percentage of remitted participants.
[c]ICER: incremental cost-effectiveness ratio ($\Delta C/\Delta E$).
[d]SA: sensitivity analysis.
[e]Sleep education with larger group size refers to having 13 participants in the SSSEI session of Trial II.



(a) (b)

FIGURE 1 Cost-effectiveness plane (A) and cost-effectiveness acceptability curve (B) of Trial II versus Trial I per percent of remitted insomnia participants.

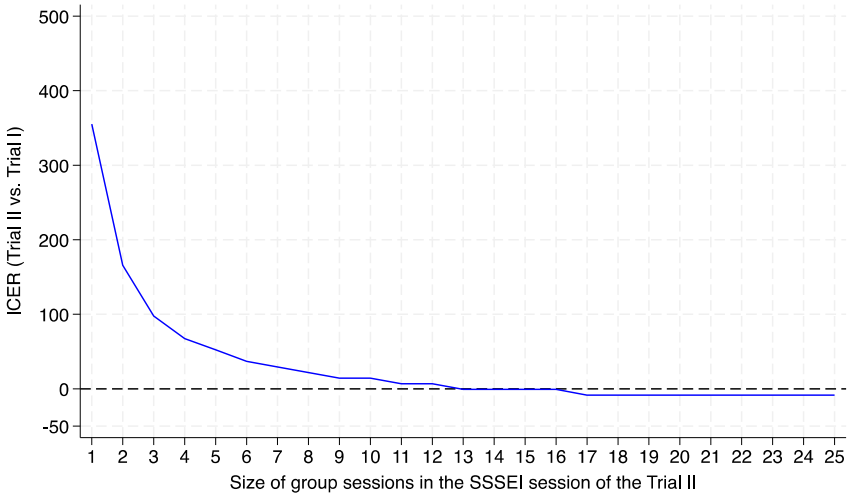


FIGURE 2 ICER of Trial II versus Trial I for various sizes of group SSSEI session of Trial II. ICER, incremental cost-effectiveness ratio; SSSEI, sessions in the single sleep education intervention.

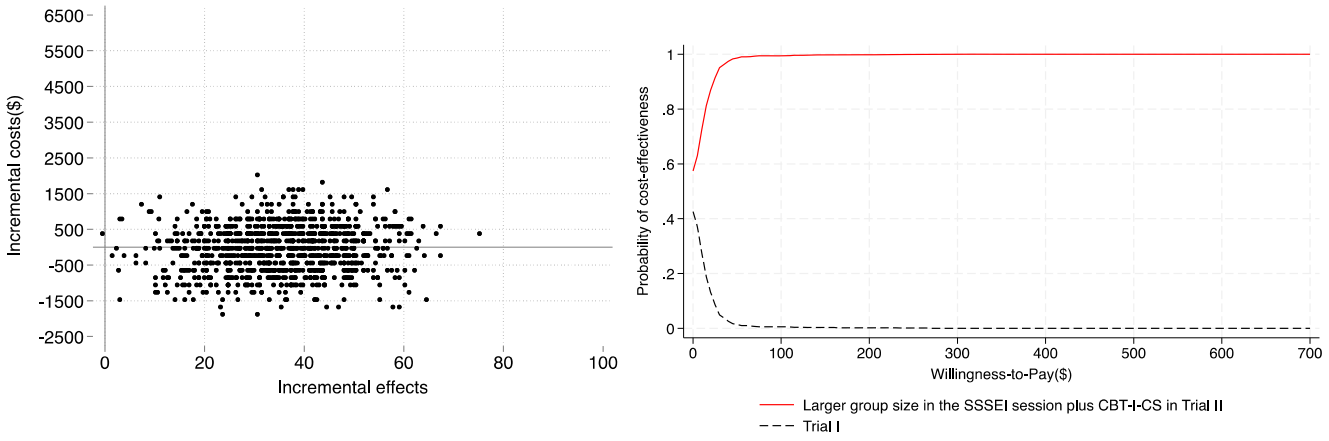


FIGURE 3 Cost-effectiveness plane (A) and cost-effectiveness acceptability curve (B) of Trial II with larger group size in SSSEI session versus Trial I. Figures illustrate the cost-effectiveness of a modified version of the Trial II, where larger group sizes were used for SSSEI session. Specifically, the larger group size consists of 13 participants per session, resulting in a total of 4 SSSEI sessions with 51 participants in the stepped care delivery of Trial II. In the actual implementation of Trial II, there were a total of 24 SSSEI sessions with an average of 2 participants per session. SSSEI, sessions in the single sleep education intervention.

4 | DISCUSSION

The cost of delivering evidence-based insomnia treatment is likely to impact the feasibility of program implementation at a cancer center. When insomnia intervention group sizes are large, the stepped care delivery approach is more cost effective than delivering care in a single treatment intervention. Specifically, a group size of 13 or more is required in order for the stepped care approach to be the economically wise choice. For cancer centers with smaller patient populations where it may be difficult to enroll a group of this size in an insomnia-focused intervention, administrators may consider providing group-based treatment alone, rather than take a stepped care approach.

4.1 | Clinical implications

There is no consensus on how stepped care models in insomnia care should be implemented. Previous stepped care insomnia treatments in the general population have included more steps than we incorporated in our trial.[20] Although these stepped care insomnia treatment models are efficient in the usage of therapeutic resources, the upper-level treatment steps can still be burdensome and face similar challenges in patient uptake and retention. Future research should study whether additional steps in the stepped care model, both at the less and more intensive ends of the spectrum, are more cost-effective than the model studied in our research. Less intensive programming may be viable, with prior research

demonstrating that video recordings can improve insomnia among cancer survivors.[21]

The natural next step of the economic evaluation of insomnia treatment for cancer survivors would be to examine the cumulative cost-effectiveness among various programs delivered across different intervention modalities and strategies, including approaches such as patient handouts, psychoeducational videos, automated Internet-based interventions, and benefits from support from clinicians or trained paraprofessionals. Furthermore, economic evaluation from multiple perspectives (e.g., patient's perspective including transportation costs, time costs, productivity loss, and other health-related costs) should be provided to guide wider program dissemination and adoption discussions.

4.2 | Study limitations

First, our data are from trials that did not have control groups. Therefore, we cannot determine the cost-effectiveness of the trials relative to no treatment. Second, the current evaluation was done from the provider's perspective only, and therefore is limited in considering patient's indirect costs and other positive and/or negative spillovers. For example, studies have shown that insomnia is associated with excessive societal costs.[22,23] Third, in this retrospective CEA, we cannot assess potential spillover costs incurred/avoided due to the CBT-I-CS treatment. However, since the stepped care and single treatment approaches are similar—both try to increase provider efficiency by using a group approach—their impact on other health care utilization, if any, should be similar. Therefore, we would expect the incremental cost-effectiveness assessment from the healthcare provider's perspective to be minimally impacted. Fourth, we were not able consider other potentially relevant factors such as tumor stage, social support and education that might impact trial outcomes in the study due to limited data availability. Last, the costs of recruitment and session organization at our institution may be different than those at other centers. A longer waiting period between groups in order to reach the minimal session size may be an important strategy to ensure treatment groups are more cost-effective.

4.3 | Conclusions

We encourage cancer center leadership to consider the feasibility of providing sleep-related care for their growing population of cancer survivors with insomnia.[24] In particular, administrators should consider the availability of local clinicians with expertise in behavioral sleep medicine and the financial implications of adding such a service. Our data suggests that programs which do not require extensive sleep expertise could be considered, potentially at a reduced cost, in the context of a stepped care model.

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CONFLICT OF INTEREST STATEMENT

Study authors do not have any conflicts of interest to declare.

DATA AVAILABILITY STATEMENT

The data supporting this manuscript can be made available upon request.

ETHICS STATEMENT

All studies were approved by the Dana-Farber Cancer Institute IRB.

PATIENT CONSENT STATEMENT

N/A.

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