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INSOMNIA

Cost-Effectiveness of Group and Internet Cognitive Behavioral Therapy for Insomnia in Adolescents: Results from a Randomized Controlled Trial

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Study Objectives: To investigate cost-effectiveness of adolescent cognitive behavioral therapy for insomnia (CBTI) in group- and Internet-delivered formats, from a societal perspective with a time horizon of 1 y

Methods: Costs and effects data up to 1-y follow-up were obtained from a randomized controlled trial (RCT) comparing Internet CBTI to face-to-face group CBTI. The study was conducted at the laboratory of the Research Institute of Child Development and Education at the University of Amsterdam, and the academic youth mental health care center UvAMinds in Amsterdam. Sixty-two participants meeting Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision criteria for insomnia were randomized to face-to-face group CBTI (GT; n = 31, age = 15.6 y ± 1.8, 71.0% girls) or individual Internet CBTI (IT; n = 31, age = 15.4 y ± 1.5, 83.9% girls). The intervention consisted of six weekly sessions and a 2-mo follow up booster-session of CBTI, consisting of psychoeducation, sleep hygiene, restriction of time in bed, stimulus control, cognitive therapy, and relaxation techniques. GT sessions were held in groups of six to eight adolescents guided by two trained sleep therapists. IT consisted of individual Internet therapy with preprogrammed content similar to GT, and guided by trained sleep therapists.

Results: Outcome measures were subjective sleep efficiency (SE) ≥ 85%, and quality-adjusted life-years (QALY). Analyses were conducted from a societal perspective. Incremental cost-effectiveness ratios (ICERs) were calculated using bootstrap sampling, and presented in cost-effectiveness planes. Primary analysis showed costs over 1 y were higher for GT but effects were similar for IT and GT. Bootstrapped ICERs demonstrated there is a high probability of IT being cost-effective compared to GT. Secondary analyses confirmed robustness of results.

Conclusions: Internet CBTI is a cost-effective treatment compared to group CBTI for adolescents, although effects were largely similar for both formats. Further studies in a clinical setting are warranted.

Clinical Trial Registration: ID: ISRCTN33922163; trial name: Effectiveness of cognitive behavioral therapy for sleeplessness in adolescents; URL: http://www.isrctn.com/ISRCTN33922163

Keywords: adolescents, cognitive behavioral therapy, cost effectiveness, insomnia, Internet therapy

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Significance

This is the first study to show that group- and Internet-delivered cognitive behavioral therapy for insomnia (CBTI) are equally effective in improving adolescent sleep, but that costs are lower in Internet-CBTI. Considering both effects and costs simultaneously, Internet-CBTI was found cost-effective compared to group-CBTI. These findings are important for policy makers and clinicians in light of the high economic burden and severe consequences of adolescent insomnia for daily life, and they highlight possibilities to disseminate CBTI more broadly via the Internet to meet the shortage of sleep therapist. Further research is recommended into effectiveness for specific subgroups (based on e.g. severity of insomnia, sex, age) and characteristics of group- and Internet CBTI.

INTRODUCTION

Insomnia disorder consists of difficulty initiating or maintaining sleep or nonrestorative sleep, accompanied by significant negative daytime consequences, for at least 3 days per week and at least 3 mo.[1] Reported prevalence of insomnia in adolescents ranges from 7.8% up to 23.8%.[2,3] Although sleep problems in children and adolescents are still severely underdiagnosed,[4] insomnia is the most prevalent sleep disorder in adolescents[5,6] and is also most often diagnosed in this age group in pediatric primary care centers.[4] Insomnia has been related to severe consequences for daily life, including anxiety and depression,[7,8] drug and alcohol abuse,[9,10] impaired academic performance,[11] problem behavior,[12] and attention deficit/hyperactivity disorder (ADHD).[13]

Over the past decade, a number of studies have been published that investigated efficacy of cognitive behavioral therapy for insomnia (CBTI) in adolescents.[9,14–19] From these studies, there is growing support for the effectiveness of CBTI for adolescents. However, because adolescents, more so than adults, appear reluctant to seek psychological help,[20] effective, low-threshold interventions need to become available that are highly accessible for this age group.[21] Internet therapy may fill this treatment gap for adolescents,[22,23] especially since the last few years mounting evidence has appeared of efficacy of pediatric online cognitive behavioral treatments compared to face-to-face treatment.[24,25] Therapist-guided face-to-face CBTI is regarded by many as the gold standard for insomnia treatment. However, the two studies on CBTI for adolescents[15,18] showed that individual guided Internet CBTI and face-to-face group CBTI were both highly feasible and acceptable for adolescents, with very low attrition rates. Moreover, there appeared no significant differences between the two treatment modalities in treating adolescents with Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision (DSM-IV-TR) primary insomnia for both short- and long-term outcomes for sleep and psychopathology up to 1 y. Considering the lack of significant differences in outcomes and the acceptability of both Internet and group CBTI, the question then remains which form of treatment would be preferable in light of other factors such as cost effectiveness.

Health care costs have risen dramatically in past decades with governments attempting to control costs and/or search for cost savings. One potential relevant source of information is cost-effectiveness studies. Cost-effectiveness studies compare both effects and costs between two (or more) treatments. Usually, an incremental cost-effectiveness ratio (ICER) is calculated, which is expressed as the difference in costs between treatments divided by the difference in effects. Next, bootstrap analyses–in which estimated ICERs are derived from repeatedly drawing random samples from the original sample–can be used to handle the uncertainty around the ICER.[26] This approach results in a cost-effectiveness plane (cross) in which the vertical axis represents the difference in costs and the horizontal axis represents the difference in effect. There are four main outcomes depending on the quadrant in which most ICERs can be found: (1) the new treatment is dominant; i.e., the new treatment is more effective and less costly (south-east quadrant), (2) the old treatment is dominant; i.e., the old treatment is more effective and less costly (north-west quadrant), (3) the new treatment is less effective as well as less costly (south-west quadrant), and (4) the new treatment is more effective as well as more costly (north-east quadrant).[27] In this latter case, the decision whether or not the new treatment is to be implemented depends on the "willingness to pay," which represents the amount of money that people (society) should be willing to pay for an extra unit of effect. One important limitation, however, is that the willingness to pay is often unknown. For example, how much money would people be willing to pay for a unit increase in sleep efficiency? One solution for this issue is to use quality-adjusted life-years (QALYs) as the outcome (effect). QALYs represent an economic measure that incorporates both the length of survival as well as the (subjective) quality of life in terms of functionality or health, and can be assessed irrespective of type of disorder or disease. ICERs can be calculated that now represent the cost per QALY. Two important advantages of using QALYs (instead of another measure of effect such as sleep efficacy) are (1) it allows comparisons between treatments across different domains, areas, disorders and/or illnesses, and (2) there is a general agreement about the willingness to pay per QALY[28] (i.e., 20,000 euro per QALY in the Netherlands[29]).

Studies on costs related to insomnia and cost-effectiveness of CBTI are scarce.[30] Results from studies with adults have shown that insomnia is associated with high direct and indirect costs.[31–33] To our knowledge, only a few studies have been published regarding the cost-effectiveness of CBTI interventions. In a review of cost-effectiveness analyses in insomnia treatment Martin et al.[34] found no studies published between 1966 and 2002, but in the discussion the authors note that the high economic burden of insomnia along with its impact on health-related quality of life (HRQOL) suggest, but do not prove, that treatment would be cost-effective. In a study comparing CBTI delivered individually, in group therapy, or through brief telephone consultations, Bastien et al.[35] concluded that because all three methods of CBTI showed similar effectiveness, telephone consultation represented a cost-effective alternative to individual or group therapy. In that study, a statistical comparison of costs and benefits between conditions was not applied and therefore these findings are inconclusive. Scott et al.[36] conducted a retrospective prevalence based cost-effectiveness analysis of insomnia treatment in New Zealand, and found a net benefit of direct costs of $482 per treated patient and an incremental net direct benefit per QALY gained when insomnia was successfully treated of $3,072. The results in their study, however, were based on cost estimates from international studies and on retrospective ratings of HRQOL of insomnia patients by two participating therapists. In a more comprehensive cost-effectiveness analysis from a health care perspective concerning psychoeducational CBTI workshops in the community, Bonin et al.[37] concluded there was a probability of 80% of cost-effectiveness at a maximum willingness to pay £30,000 per QALY. At a willingness to pay the net cost of the intervention of £150 per point improvement on the Insomnia Severity Index,[38] they found a 97% probability of cost-effectiveness. This analysis, however, used only health-care service costs to analyze differences between participants who did or did not participate in the CBTI workshops, and furthermore, the workshops were compared to a waiting list condition, which was likely to result in cost-effectiveness of the workshops, as cost of illness of insomnia is high.[34] Finally, in a cost-effectiveness analysis of treatment as usual (TAU) with or without additional CBTI for 37 patients with major depressive disorder and comorbid insomnia, Watanabe et al.[39] found that with a willingness to pay $40K more for one more QALY, TAU plus CBTI would be cost effective with a probability of 90% compared to TAU alone. Note that none of these studies was conducted with adolescents, and none of these studies provided a complete cost-effectiveness analysis of CBTI only, based on data from a randomized clinical trial.

In contrast to the paucity of cost-effectiveness studies on CBTI, in the relatively young and growing field of behavioral Internet interventions, a number of studies on cost-effectiveness analyses have recently been published. In a review from 2009, Tate et al.[40] found eight studies that reported specific economic indicators of Internet interventions, although most of these studies lacked comprehensive analyses, and the authors concluded that it is imperative for this emerging field to capture and report cost effectiveness. Five years later in a systematic review of Internet delivered treatments for mood and anxiety disorders Arnberg et al.[41] found five studies that reported cost effectiveness but excluded three due to high risk of bias. The authors concluded that the quality of evidence for cost-effectiveness was low/very low. One study from that review compared costs and effects between Internet and group treatment for social anxiety disorder.[42] The Internet condition showed a lower cost per patient of $1,422 and 19% greater improvement of outcomes at 6-mo follow-up. At a willingness to pay $3,000 per additionally improved patient, there was a probability of 90% that Internet treatment was cost effective compared to group treatment. In a 4-y follow-up of that study,[43] the authors found similar effectiveness of both treatments, and only a very slight difference in cost effectiveness, indicating that with a willingness to pay $0 the Internet treatment had a probability of 62% of being more cost effective than the group -treatment. This dropped to 22% if willingness to pay was increased to $100,000. None of these studies on cost effectiveness of Internet interventions reported on Internet CBTI in general, or any psychological treatment for adolescents specifically.

In summary, research indicates that CBTI in Internet- and face-to-face group formats can be effective in the treatment of insomnia in adolescents, and Internet-delivered CBTI could both lower the threshold for adolescents to seek treatment, and provide a cost-effective alternative for face-to-face group therapy. However, research into cost-effectiveness of CBTI for adolescents is lacking, and additionally, there is a lack of cost-effectiveness analyses of Internet interventions for adolescents.

In the current study we therefore investigated cost-effectiveness of CBTI delivered through the Internet (IT) compared to CBTI in face-to-face group therapy (GT). Sixty-two adolescents were randomly assigned to GT or IT. Societal costs, insomnia outcomes, and HRQOL were measured at baseline, at posttreatment, and 1-y follow-up. We hypothesized that IT would be cost effective compared to GT.

Table 1—Demographic characteristics of the participants.

Characteristic	IT (n = 31)	GT (n = 31)
Age (mean, SD) in y	15.4 (1.5)	15.6 (1.8)
Sex, n (%)		
Female	26 (83.9)	22 (71.0)
Male	5 (16.1)	9 (29.0)
Parent marital status, n (%)		
Married or living together	28 (90.3)	22 (71.0)
Single	0 (0.0)	4 (12.9)
Living apart together	2 (6.5)	3 (9.7)
Other	1 (3.2)	0 (0.0)
Missing	0 (0.0)	2 (6.5)
Parent education, n (%)		
High school or less	1 (3.2)	0 (0.0)
Some college	10 (32.3)	12 (38.7)
College graduate	9 (29.0)	7 (22.6)
Graduate school	10 (32.3)	10 (32.3)
Other	1 (3.2)	0 (0.0)
Missing	0 (0.0)	2 (6.5)
Parent occupational status, n (%)		
Working 75–100%	18 (58.1)	11 (35.5)
Part time	11 (35.5)	13 (41.9)
Other	2 (6.5)	5 (16.1)
Missing	0 (0.0)	2 (6.5)
Parent nationality, n (%)		
Dutch	31 (100)	28 (90.3)
Other	0 (0.0)	1 (3.2)
Missing	0 (0.0)	2 (6.5)

GT, group therapy; IT, internet therapy.

METHODS

Study Design

This study was a prospective cost-effectiveness analysis from a societal perspective with a time horizon of 1 y. In a parallel group, randomized design, participants were assigned to GT or IT using simple randomization, with an equal allocation ratio, by referring to a table of random numbers. The trial followed CONSORT 2010 guidelines,[44] for reporting parallel group randomized trials, recommendations on measuring outcomes in insomnia trials,[45] and the Dutch Guideline for Cost Research.[46] This study was part of a larger randomized controlled trial, which was approved by the medical ethics committee of the Academic Medical Centre in Amsterdam and registered at http://www.isrctn.com/ (ISRCTN33922163). The main outcome study has been reported elsewhere.[18]

Participants

The participants were recruited with newspaper articles, electronic newsletters, and leaflets that were spread among healthcare practitioners and secondary schools within a 50-km radius from the research and treatment facilities. In total 478 participants were screened online, of who 342 left the study or were excluded based on inclusion and exclusion criteria. Inclusion criteria were (1) age between 12–19 y, (2) in secondary school or after (i.e., further education or work), (3) living within traveling distance, and (4) meeting the diagnostic criteria of the DSM-IV-TR for primary insomnia. These criteria concern difficulties falling or staying asleep, or not feeling rested after getting up, presence of these problems for at least 1 mo, and clinically significant consequences for daily life.[47] Exclusion criteria were other sleep problems, other psychiatric problems (including suicidal plans), physical problems that could interfere with sleep, and drug or medication use (including melatonin) that could affect sleep. Retrospective analysis of the intake data of the participants revealed that all participants also met the DSM-5 criteria for insomnia disorder, which include that the problems are present for at least 3 mo, and at least 3 days per week.[1] Initially 39 participants participated in IT, and 38 in GT. Due to a technical issue with the user settings of the online cost questionnaires, the first 8 participants from the IT condition and the first 7 from the group condition did not automatically receive the cost questionnaires and therefore did not participate in any of the cost measurements. Therefore, in the current study, 31 participants participated in GT, and 31 participants participated in IT (see Figure 1 and Table 1). The 31 participants in the IT condition who participated in this study did not differ in age ($t(37) = 0.36$, $P = 0.724$) or sex ($X^2(1) = 0.06$, $P = 0.800$) from the 8 who did not participate. The 31 participants in the GT condition who took part in this study did not differ in age ($t(36) = 0.57$, $P = 0.571$) or sex ($X^2(1) = 0.51$, $P = 0.477$) from the 7 who did not participate. The participants who did not participate did not differ between GT and IT in age ($t(13) = 1.06$, $P = 0.309$) or sex (Fisher exact test $X^2(1) = 1.76$, $P = 0.282$).

Interventions

CBTI consists of a set of techniques that have been developed for treatment of insomnia in adults. For the current study the treatment protocol was adapted from CBTI for adults, with age-appropriate exercises for adolescents, by the research

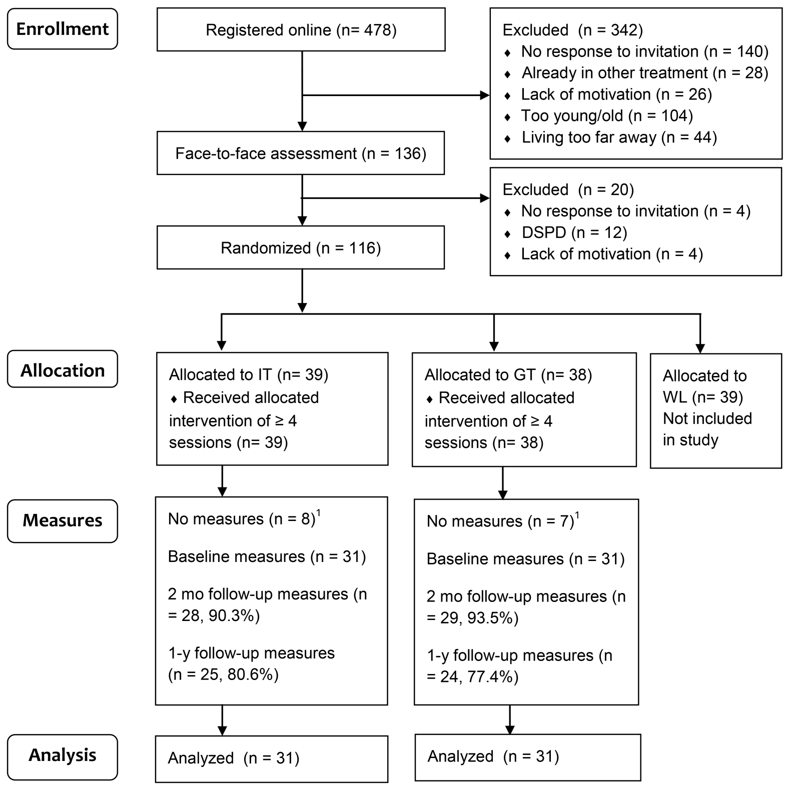

Figure 1—Participant flow through the recruitment and research procedure. [1]No measures of 8 participants in IT and 7 in GT due to a technical issue with the cost-questionnaires. DSPD, delayed sleep phase disorder; GT, group therapy; IT, Internet therapy; WL, waiting list.

team and an experienced sleep specialist. It contained sleep hygiene, psychoeducation, restriction of time in bed, stimulus control, cognitive therapy, and relaxation techniques, ordered over six weekly consultations in a similar fashion for both IT and GT. After 2 mo there was a booster session with a short overview of all exercises from the previous six sessions. Participants who were assigned to GT came to a local mental health care center for youth and their parents, where they received 1.5-h sessions with two trained sleep psychotherapists in groups of six to eight participants. They received a binder in which to save all leaflets and printed homework exercises, which they brought to all sessions. Participants in the IT condition logged onto a personal login page of a website where at a fixed day and time each week a session was made available. Each participant in IT was guided by a personal sleep-psychotherapist for the duration of the treatment. Before each session, except the first, participants in IT answered questions on how well exercises went over the past week. Automated feedback related to the answers to these questions was embedded in the pages for each consult. Furthermore, for each session the personal therapist wrote a short personalized feedback on data from the sleep logs that participants filled out each day, and gave personal bedtimes-advice. This feedback consisted of 352.0 words on average (standard deviation = 104.6). After the first session participants received an invitation for a 15-min chat session with their therapist that took place in the week after the second session. De Bruin et al.[15] provide a more detailed description of both treatment modalities.

Measures

For comparisons of cost effectiveness we conducted a primary analysis and several secondary analyses to test robustness of the outcomes from the primary analysis. Measures were obtained at baseline, at posttreatment after the 6 w of treatment were completed but before the booster session (i.e. approximately 15 w after baseline), and at 1-y follow-up (i.e., approximately 37 w after posttreatment).

Outcome Assessment

As a proxy for recovery subjective sleep efficiency (SE) from sleep logs was used. SE is regarded as a reliable and valid outcome measure of insomnia treatment, as it catches both the difficulty with initiating and maintaining sleep. A score of SE ≥ 85% is regarded as sufficient. The sleep log was based on the consensus sleep diary[48] and consisted of eight questions concerning bedtime, time of lights out, sleep onset latency (SOL), number and time of wake after sleep onset (WASO), wakeup time, get-up time, and subjective sleep quality. Participants filled out the sleep logs within 1 h after getting up, for 7 consecutive days at each measurement occasion. Sleep logs could be filled out up to midnight the following day, at the latest, as retrospective data with a larger time span were considered unreliable. SE consists of the percentage total sleep time (TST) from the time spent in bed (TIB). TIB was calculated by the difference of "time of lights out" and "get-up time." TST was TIB minus SOL, minus time of WASO, and minus time between "wake-up time" and "get-up time."

To calculate QALY scores the quality of life (QOL) of the participants was rated using the EuroQol-5D (EQ-5D[49]), which has good psychometric properties.[50–52] The questionnaire contains five dimensions of HRQOL (i.e., mobility, self-care, usual activities, pain/discomfort, anxiety/depression) for which the participants rated whether they had no, some, or severe problems. A health state index (utility score) was calculated by attaching preference weights to each dimension.[53] QALYs were calculated by taking the average utility score from baseline and posttreatment measures, and posttreatment and 1-y follow-up measures, multiplying these averages by the fraction of the year that the time between the measures represented, and adding them. The time-horizon of the comparisons is 1 y and therefore the maximum QALY is 1.

Cost Assessment

At each measurement occasion, parents filled out retrospective cost questionnaires that reported on resource usage over the past 2 mo (e.g., doctors' visits, use of medication, mental health care visits, additional help at school/home, etc.). The items of the cost questionnaire were based on the cost diary used in other studies[54,55] and adapted for use in this study. A family perspective was used, meaning all costs related to the adolescent were taken into account, including direct and indirect costs for health care usage such as doctor visits and medication use, and direct and indirect non-health care costs such as informal care, parents' loss of (non)paid work, traveling expenses, and tutoring of the adolescent. Costs were calculated by multiplying the resources used by the unit price of each resource. Unit prices were obtained from the Dutch Guideline for Cost Research,[46] the Dutch Healthcare Authority, the Dutch government, and the Centraal Plan Bureau (CPB) Netherlands Bureau for Economics Policy Analysis. For the costs of medications, information was retrieved from the Dutch website of the "Pharmaceutical Compass" (and for a few over-the-counter medications, an online drugstore was consulted; Drogisterij.net[56]). Shadow prices were used if an official price unit was not available and the friction cost method was used to calculate productivity losses of parents.[46] All costs were indexed at 2014 euro based on Dutch price-index values.[57] See Table 2 for an overview of unit prices.

Cost of Internet and Group CBTI

Costs of CBTI were calculated based on the hours spent by therapists to apply the CBTI protocol. For both therapy modalities, therapists registered the hours spent to prepare and deliver the consults, for administrative purposes, and for intervision and supervision sessions.

Analyses

At posttreatment, measures were provided by 29 participants from GT and by 28 participants from IT. At 1-y follow-up, 24 in GT and 25 in IT provided measures. There was 12% missing data in our sample. The Little Missing Completely at Random test was not significant ($X^2(1{,}128) = 1{,}070.79$, P = 0.887), which indicates that these data were missing completely at random. Therefore, incomplete data were imputed at the item level using SPSS 22.0 (IBM corporation, NY, USA) multiple imputations.[58] Chi-square tests were used to analyze differences of proportions of participants with sleep efficiency (SE) ≥ 85%. To examine differences between IT and GT at baseline of mean QALYs, Mann-Whitney *U* tests were used. Differences of societal costs and health care costs were examined with bootstrap analyses (n = 1,000). Cost effectiveness was analyzed using bootstrapped (n = 1,000) estimated ICERs. Bootstrap analyses draws, with replacement, a number of samples from the original data to estimate the sampling distribution and its 95% confidence interval, and to quantify the uncertainty around the costs and cost-effectiveness ratios.[26] ICERs were expressed as the cost per participant reaching SE ≥ 85% (as a proxy for recovery), and as the cost per QALY. Cost-effectiveness planes were used to represent the bootstrapped ICERs, with the vertical line reflecting the difference in costs and the horizontal line reflecting the difference in effect between conditions. Cost-effectiveness acceptability curves were used to represent the probability that the compared treatment (IT compared to GT) is cost effective, with a range of willingness-to-pay ratios that depict costs for one additional adolescent with SE ≥ 85%, or one additional QALY, in order for the treatment to become cost effective.

Table 2—Unit price of each resource (indexed at 2014 euros).

Resource	Unit Price (2014 euros)
General practitioner	
Visit	30.78 p/c
Home-visit	47.26 p/c
Telephone	15.39 p/c
Polyclinic visit	141.79 p/c
Mental health care	
Primary psychologist	87.93 p/c
Social worker	71.45 p/c
Psychiatrist	113.21 p/c
Psychotherapist	84.64 p/c
Paramedic care	
Physiotherapist	39.57 p/c
Other paramedic care	34.07 p/c
Medication	
Prescription	0.12–1.29 p/dose
Without prescription	0.22–0.82 p/dose
Indirect costs	
School absenteeism	5.30 p/h
Tutoring	21.71 p/h
Loss of daily activities adolescent	5.30 p/h
Loss of paid work parents	32.12 p/h
Loss of daily activities parents	13.78 p/h
Travel expenses	
Car/public transport	0.22 p/km
Parking costs	3.27 p/o
Costs group therapy	467.75 total
Costs Internet therapy	396.62 total

All costs in 2014 euro. Index based on Dutch price index values.[57] p/c, per contact; p/dose, per dose (tablet, drops, etc.); p/h, per hour; p/km, per kilometer; p/o, per occasion.

Secondary Analyses

To test robustness of the primary analyses, several secondary analyses were performed with alternative parameters to calculate cost-effectiveness. First we used only health care-related costs instead of societal costs. Second, instead of SE ≥ 85% we used the reliable change index (RCI) of SE[59] as measure of effect. Third, we used as alternative effect measure the RCI of SOL of the sleep logs, as a high SOL is often the predominant complaint of adolescents.[60] Fourth, we used the insomnia scale from the Holland Sleep Disorder Questionnaire (HSDQ[61]) as effect measure. The insomnia scale of the HSDQ (HSDQi) consists of eight items, scored on a five-point rating scale. Good psychometric properties have been demonstrated, and a score above the cutoff of 3.167 on a range of 1 to 5 on the insomnia scale is an indication of insomnia for adolescents. Finally, we incorporated the ongoing costs of Internet CBTI in the total societal costs for IT. We based the calculations of ongoing costs on the actual costs that were incurred during the 3 y that the participants in this study were treated with Internet CBTI, divided by the total amount of participants who were treated with Internet CBTI. These costs were hosting

Table 3—Quality of life, sleep efficiency, societal and health care costs, and secondary measures at baseline, posttreatment, and at follow-up.

Category	IT (n = 31)	GT (n = 31)	Difference (IT-GT)		P
Primary analyses					
Sleep efficiency ≥ 85%				$X^2(1)$	
Baseline	36%	29%	7%	0.30	0.587
Posttreatment	87%	74%	13%	1.65	0.199
Follow-up	90%	94%	−4%	0.22	0.641
Quality of life	Mean (SD)	Mean (SD)		U [c]	
Baseline	0.83 (0.20)	0.86 (0.15)	−0.03	469.0	0.867
Posttreatment	0.88 (0.16)	0.88 (0.17)	0.00	445.5	0.603
Follow-up	0.93 (0.07)	0.91 (0.13)	0.02	465.5	0.825
QALYs over 3 mo	0.25 (0.05)	0.25 (0.04)	0.00	452.0	0.680
QALY's over 12 mo	0.89 (0.11)	0.89 (0.12)	0.00	475.5	0.943
Costs for CBTI [a]	396.62	467.75	−71.13		
Societal costs [a]					
Baseline	494.83 (879.25)	472.68 (678.33)	22.15	444.5	0.612
Posttreatment	747.76 (253.41)	877.42 (142.65)	−129.66	126.0	< 0.001
Follow-up	298.77 (579.75)	574.66 (1,487.92)	−275.89	387.0	0.176
Total societal costs over 12 mo [a]	1,046.53 (696.03)	1,452.08 (1,491.26)	−405.55	285.0	0.006
Secondary analyses					
Health care costs [a]					
Baseline	75.92 (150.36)	38.48 (76.64)	37.44	413.0	0.289
Posttreatment	650.66 (112.88)	799.83 (52.70)	−149.17	29.0	< 0.001
Follow-up	38.57 (79.69)	36.23 (71.32)	2.34	415.5	0.331
Total health care costs over 12 mo [a]	689.23 (138.91)	836.06 (107.75)	−146.83	118.0	< 0.001
				$X^2(1)$	
RCI HSDQi [b]	45%	52%	−7%	0.26	0.611
RCI sleep efficiency [b]	23%	48%	25%	4.51	0.034
RCI SOL [b]	58%	58%	0%	0.00	0.601

[a] Costs in euros. Costs in IT and GT at posttreatment include costs for CBTI. [b] Reliable change index[59] for difference between baseline and 12 mo follow-up. [c] Mann-Whitney *U* test. CBTI, cognitive behavioral therapy for insomnia; GT, group therapy; HSDQi, Holland Sleep Disorder Questionnaire insomnia scale; IT, Internet therapy; QALY, quality-adjusted life-years; RCI, reliable change index; SOL, sleep onset latency.

costs, costs for secure servers, and irregular maintenance costs, which in total amounted to €295.50 per participant, increasing the total costs of Internet CBTI to €692.12. The "sunk costs" (i.e., the costs to have the Internet CBTI application built) were not used in these additional cost calculations, because from an economical perspective these costs will not be repeated.[40]

RESULTS

Table 1 shows demographic characteristics of the participants. There were no differences in age ($t(60) = −0.46$, P = 0.650) or sex ($X^2(1) = 1.48$, P = 0.363) between these groups. No significant differences were found between the groups at baseline for proportions of participants with SE ≥ 85% ($X^2(1) = 0.30$, P = 0.587) and QOL (Mann-Whitney *U* test = 469.00, $Z = −0.17$, P = 0.867). Bootstrap analyses revealed that at baseline there were no significant differences between IT and GT of total societal costs (mean incremental costs = €82, 95% CI = -€367 to €578) or health care costs (mean incremental costs = €65, 95% CI = €3 to €140). See Table 3 for the scores of SE ≥ 85%, QOL, QALY, societal costs, and health care costs at baseline, posttreatment, and 1-y follow-up, and secondary outcome measures for the two conditions.

Primary Analyses of Cost-Effectiveness of Internet CBTI Compared to Group CBTI

At 1-y follow-up, there was no significant difference in proportions of participants in IT and GT who had SE ≥ 85%, (90% and 94% for IT and GT, respectively, $X^2(1) = 0.22$, P = 0.641). At 1-y follow-up there appeared no differences between IT and GT for mean QALY (0.89 for both IT and GT, Mann-Whitney *U* test = 475.50, $Z = −0.07$, P = 0.943). Although there were differences between IT and GT of total societal and health care costs at 1-y follow-up, mainly due to different CBTI costs of €396.62 for IT compared to €467.75 for GT, bootstrap analyses revealed that these differences were not significant.

Table 4 shows costs, effects, and ICERs for IT versus GT. The ICER based on societal costs per adolescent with SE ≥ 85% demonstrated that costs are lower for IT, but effects are slightly higher for GT, with one more participant in GT (4%) reaching SE ≥ 85%. The cost-effectiveness plane based on bootstrapped ICERs for total societal costs and participants with SE ≥ 85%

Table 4—Effects, costs, and cost-effectiveness analyses for Internet CBTI (IT) versus group CBTI (GT).

	IT		GT		ICER	% ICER falling into the four quadrants of the cost-effectiveness plane				Probability (%) that IT is cost-effective at willingness to pay of (€)			
	Cost	Effect	Cost	Effect		NE	NW	SW	SE	0	5,000	10,000	20,000
Primary analysis													
Recovery (SE ≥ 85%)	1,047	0.90	1,452	0.94	12,572	2	3	58	37	95	71	54	42
QALY	1,047	0.89	1,452	0.89	Dominant	2	4	44	50	95	91	86	76
Secondary analyses													
Health care perspective	689	0.90	836	0.94	4,552	0	0	58	42	100	49	42	42
RCI sleep efficiency	1,047	0.23	1,452	0.48	1,571	0	4	95	2	97	8	4	2
RCI sleep onset latency	1,047	0.58	1,452	0.58	Dominant	3	2	43	52	95	75	63	56
HSDQi recovery	1,047	0.45	1,452	0.52	6,286	0	5	60	35	95	55	43	37
Including ongoing costs	1,342	0.90	1,452	0.94	3,412	11	29	37	24	61	47	40	35

GT, group therapy; HSDQi, Holland Sleep Disorder Questionnaire insomnia scale; ICER, incremental cost-effectiveness ratio; IT, Internet therapy; NE, North-East (IT > effect and > costs than GT); NW, North-West (IT < effect and > cost than GT); QALY, quality-adjusted life-years; RCI, reliable change index[59] for difference between baseline and 12-mo follow-up; SE, sleep efficiency; SOL, sleep onset latency; SW, South-West (IT < effect and < costs than GT); SE, South-East (IT > effect and < costs than GT); SE ≥ 85%, sleep efficiency from sleep logs ≥ 85% at 1-y follow-up.

showed most point estimates falling in the south side of the plane (95%), indicating that overall there is a high probability of IT being less costly than GT. The somewhat higher percentage of point estimates on the west side of the plane (61%) indicates a slightly lower effect for IT.

The ICER based on societal costs and QALYs demonstrated that IT dominates GT. The percentage of point estimates on the south side of the plane (94%) indicate lower cost for IT compared to GT, and the percentage on the east side of the plane (52%) indicate similar effects for IT and GT (Figure 2).

The cost-effectiveness acceptability curves (Figure 3) show that with a willingness to pay €0, the probability of IT to be cost effective is 95% for both SE ≥ 85%, and QALYs. If willingness to pay would increase to €20,000, the probability of cost-effectiveness of IT would decrease to 42% for SE ≥ 85%, and to 76% for QALYs.

Secondary Analyses

Analyses of ICERs from a health care perspective including only health care costs, the RCI of SOL, and the RCI of the HSDQi showed comparable results to the primary analyses (i.e., effects being more or less the same for IT and GT, with costs being lower in IT). See Table 4 for ICERs and probabilities of cost-effectiveness of IT at different levels of willingness to pay for these scenarios.

For two scenarios in the secondary analyses, the results were somewhat different. First, using the RCI of SE as effect measure showed a distinctly lower effect for IT (23% was meaningfully improved in IT versus 48% in GT) at the same societal costs as in the primary analysis (i.e., lower costs for IT). At willingness to pay of €0, there is a 100% probability of cost-effectiveness of IT compared to GT, but this decreases rapidly if willingness to pay is increased, with a probability of cost-effectiveness for IT of 2% if willingness to pay increased to €20,000. Second, using the main outcome measure SE ≥ 85% for effect, but including ongoing costs for Internet CBTI, the difference of total societal costs between IT and GT becomes much smaller and therefore the probability of IT being less costly is lower (60% of point estimates on the south side of the cost-effectiveness plane). The probability of IT being cost effective at a willingness to pay of €0 decreases from 95% (IT without ongoing costs) to 61% (IT including ongoing costs). The probability of IT including ongoing costs being cost effective decreases to 35% if willingness to pay increases to €20,000.

DISCUSSION

This was the first study to compare cost-effectiveness of Internet CBTI to group CBTI for adolescents. Although the effects were slightly better for group CBTI in most of the effect measures used in the primary and secondary analyses, Internet CBTI had considerably lower costs than group CBTI, mainly due to lower costs for the treatment, and therefore had a high chance of being cost effective. With a willingness to pay extra for additional effect, the cost-effectiveness of IT compared to GT decreased.

In this cost-effectiveness study we compared Internet CBTI to traditional face-to-face group CBTI for adolescents, wherein the face-to-face group CBTI acts as the TAU condition, or gold standard, which is used to compare alternative treatments to in cost-effectiveness studies. However, both delivery modalities for CBTI in this study were completely novel treatments, following a similar protocol for CBTI. Because behavioral therapy has been the first choice of treatment for insomnia for over a decade,[62,63] unfortunately there is a shortage of sleep therapists, and in the Netherlands most people with insomnia complaints consult their general practitioner.[64] As a consequence, TAU for adolescents with insomnia most likely consists of a form of individual therapy with elements from CBTI and medication. In this respect, it has to be noted that a large group of adolescents do not receive any treatment, and because spontaneous recovery from insomnia is not likely,[5] both types of delivery modalities for CBTI used in the current study may be likely candidates for cost-effective interventions compared to real TAU. In the intake interviews most participants mentioned

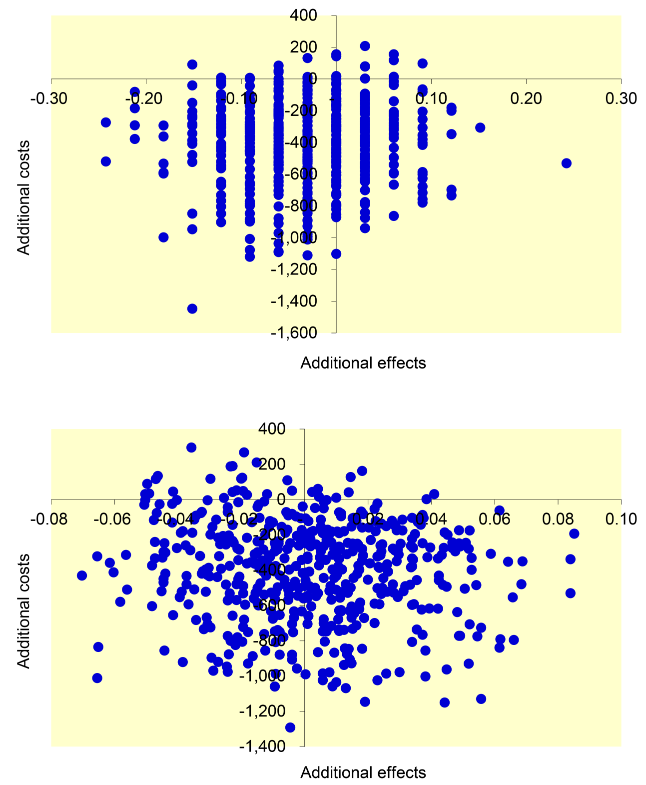

Figure 2—Cost-effectiveness planes of Internet cognitive behavioral therapy for insomnia (CBTI) versus group CBTI for total societal costs from baseline to 1-y follow-up, and effects on percentage of participants with sleep efficiency ≥ 85% as a proxy for recovery (top panel), and quality-adjusted life-years (bottom panel).

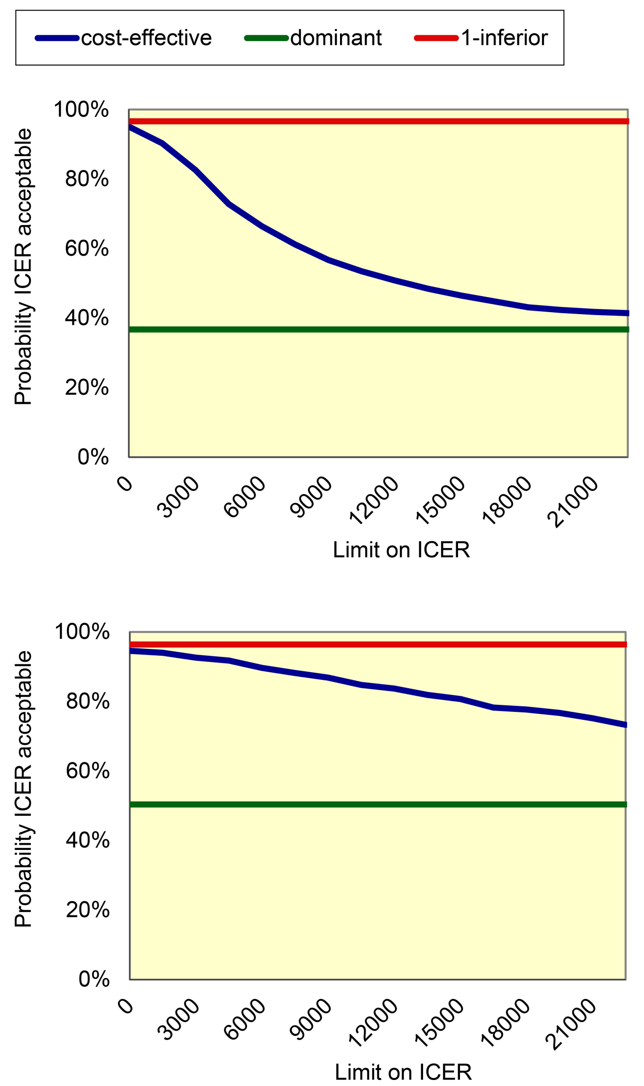

Figure 3—Cost-effectiveness acceptability curves of Internet cognitive behavioral therapy for insomnia (CBTI) versus group CBTI for total societal costs from baseline to 1-y follow-up, and effects on percentage of participants with sleep efficiency ≥ 85% as a proxy for recovery (top panel), and quality-adjusted life-years (bottom panel). ICER, incremental cost-effectiveness ratio.

they had consulted health care professionals before enrolling in our study, and had received some advice on sleep hygiene practices, or had used medication such as melatonin, or other over-the-counter drugs, without resulting in the desired relief from insomnia. This indicates that TAU for them had not been effective. From the baseline measures there appeared to be a societal cost over the course of the previous 2 mo, on average €484. Extrapolated over 12 mo, this would indicate a societal cost of €2,904 per year for each adolescent with insufficient treatment of insomnia. The total societal costs over 12 mo of our groups amounted to €1,047 and €1,452, which are both well below €2,904, and indicates that either treatment would be cost effective compared to the TAU our participants received before enrolling in our study.

In six of the seven analyses we conducted, IT had a very high probability, between 95% and 100%, to be cost effective compared to GT if willingness to pay would be €0. However, in all scenarios there was a slightly smaller effect for IT, with the largest difference between IT and GT for the RCI of SE, and the analyses showed that with an increasing willingness to pay the probability of IT being cost effective decreased. However, the reason for the large difference in proportion of participants for the RCI of SE could be that the participants in IT started out at baseline with a slightly better SE, which left less room for improvement. This is further confirmed by the fact that almost the same proportions of participants in both conditions reached SE ≥ 85% (i.e., 90% and 94% for IT and GT, respectively). Internet CBTI could therefore function as the first choice of treatment delivery for an adolescent in a stepped care model for insomnia,[65] with group CBTI as an alternative or secondary choice. Moreover, because the acceptability of Internet CBTI is high among adolescents,[15,22,23] this could fill the gap that currently exists in demand for treatment. This is even more important considering the negative consequences of insomnia and the subsequent chronic sleep reduction it causes, such as higher risk for other psychological and somatic disorders and their related costs.[7–13,27–29] Further studies into cost effectiveness of GT and CBTI for adolescents with more resistant types of insomnia are warranted.

In one of our secondary analyses we incorporated ongoing costs for Internet CBTI to gain some perspective if Internet CBTI would turn out to be more costly than traditionally thought, which we based on actual costs incurred during this study. The results from that analysis showed a probability of IT being cost effective that was distinctly lower than without ongoing costs included for IT (61% versus 95%). However, these ongoing costs may have been overestimated, because we treated only a limited amount of participants, whereas in clinical practice the capacity of the Internet CBTI application would allow for many more adolescents to be treated, thus lowering the average ongoing cost per adolescent. Furthermore, we calculated therapy costs based on actual time spent by the therapists on the treatment. Ultimately the cost of treatment would be fixed in a set fee per patient, which has not been established for online CBTI in the Netherlands yet, although for the purpose of calculating cost-effectiveness based on actual cost differences it stands to reason to use actual time spent. However, we did not include sunk costs for Internet CBTI. This type of cost in traditional face-to-face therapy can for instance represent the costs for the facilities where the treatment is delivered, and such costs will only be incurred once. The sunk costs for a web application such as our Internet CBTI application, however, would have to be provided more often as developments in Internet technology are still occurring quickly, and regular updates for layout, technological innovations, and possibly even incorporating new and additional therapeutic modules, could be needed to stay current (e.g., use of social media, use of video conferencing, use of mobile technology such as smartphones). There is no research available about the maximum lifespan of e-health applications, although for general websites it is usually estimated between 3 and 5 y, and after that period technical and design updates would be necessary. The area of Internet delivered psychological treatment is still in its early stages, and effectiveness and cost-effectiveness studies have only started to appear since roughly one decade. Further studies into the lifespan of Internet treatments are needed, and which costs to regard as ongoing costs and sunk costs.

From cost-of-illness studies with adults it is clear that insomnia is associated with very high societal costs.[31–33] According to Daley et al.[31] the average annual per-person cost (direct and indirect combined) was $5,010 for individuals with insomnia syndrome, and $1,431 for individuals with symptoms of insomnia, compared to $421 for good sleepers. Furthermore, insomnia is related to psychopathology[9,11–13] and there is increasing evidence that insomnia can contribute to or cause other mental disorders,[8,19] which in turn impose an extra economic burden on society. Insomnia in adolescents has been shown to be highly chronic,[5] so adolescents with untreated insomnia are likely to become adults with insomnia. In addition to the effect that insomnia has on QOL,[66] these considerations of economic burden and related psychopathology underscore the importance to identify insomnia as early as possible. The high feasibility, accessibility, and largely similar effects of IT compared to GT, and the high probability of IT being cost effective compared to GT at a willingness to pay of €0 that is demonstrated in this study, all are arguments to disseminate CBTI for adolescents on a wide scale, which could be facilitated through the Internet.

For both modalities the treatment costs were relatively low. Although in clinical practice there might be a reduced productivity, and thus higher CBTI treatment costs, due to the "selection bias" problem, which purports that the choice/quantity of mental health care may be correlated with unobserved variables that lead to a bias in the estimate of the effect of care,[67] or due to poorer treatment planning as modeled in what is known as the Little Law,[68] the intervention costs in this study are well below the annual costs of $5,010 mentioned in cost of illness studies for adults.[31]

Despite its merits, such as being the first cost-effectiveness study on treatments for adolescents with insomnia, and to our knowledge also the first study on cost effectiveness of an Internet mental health intervention for adolescents, this study has some limitations. First, the group CBTI that we used to compare with IT was a novel treatment itself. TAU would be the health care that adolescents would have received prior to our study, which in some cases would have been Group-CBTI, but in most cases would be use of over-the-counter medication, and sleep hygiene advice from health care professionals.[64] Because we have not used data from TAU in our study, the cost effectiveness of GT in comparison with TAU was not established, although research on economic burden of insomnia indicates a high probability of CBTI to be cost effective,[34] and as mentioned before, compared to the measurements of costs before enrolling in our study, the large decrease of costs after treatment indicates that both GT and IT would be cost effective compared to TAU. The second limitation of this study concerns the estimates of costs for Internet treatment. As we described before, there are some uncertainties about which costs to regard as ongoing and which are sunk costs. The additional €295.50 for CBTI in our secondary analyses might be overestimated if the number of treatments with Internet CBTI increased. However, it could also be underestimated if other costs such as regular technological and design updates would be included. Ultimately, the costs of the treatment should result in a fixed fee per patient, which would inform policy makers on health care costs if CBTI were to be disseminated on a wider scale. For these decisions a clear insight into sunk costs, ongoing costs and per patient fees, especially for Internet CBTI, are needed. Further research into this issue is warranted. A third limitation could be the sample size of our study. Although we recruited a decent sample size for this study, a technical complication somewhat reduced the groups that we could include in the analysis. In comparison with other cost-effectiveness studies our sample is modest to average,[39,41] but six of the seven analyses we conducted showed a very high probability of IT to be cost effective compared to GT, and therefore the results seem robust. A fourth limitation of our study is that we conducted analyses based on intent-to-treat, but not a per-protocol analysis, and therefore conclusions about noninferiority of the two treatment modalities cannot be formulated. Although there appeared to be no significant differences in effectiveness in our study, further research into CBTI treatments based on noninferiority analyses is warranted.

In this study we aimed to investigate cost effectiveness of IT compared to GT, hypothesizing that IT would be more cost effective. One of the aims of the main outcome study (a randomized controlled trial including IT and GT compared with a waiting list condition)[18] was to develop a low-threshold, clinically effective, and cost-effective treatment for adolescents with insomnia. The results from this study, indicating that IT is cost effective compared with GT, confirm that this aim is met. To conclude, as insomnia appears widespread among adolescents, and has severe consequences for psychopathology, functioning in daily life, and a high subsequent economic burden, further dissemination of CBTI for adolescents is warranted, accompanied by further research into effectiveness of CBTI in a clinical setting, and costs and effects on related psychopathology.

REFERENCES

1. American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders, 5th ed. Arlington, VA: American Psychiatric Association, 2013.
2. Dohnt H, Gradisar M, Short MA. Insomnia and its symptoms in adolescents: comparing DSM-IV and ICSD-II diagnostic criteria. J Clin Sleep Med 2012;8:295–9.
3. Hysing M, Pallesen S, Stormark KM, Lundervold AJ, Sivertsen B. Sleep patterns and insomnia among adolescents: a population-based study. J Sleep Res 2013;22:549–56.
4. Meltzer LJ, Johnson C, Crosette J, Ramos M, Mindell JA. Prevalence of diagnosed sleep disorders in pediatric primary care practices. Pediatrics 2010;125:E1410–8.
5. Johnson EO, Roth T, Schultz L, Breslau N. Epidemiology of DSM-IV insomnia in adolescence: lifetime prevalence, chronicity, and an emergent gender difference. Pediatrics 2006;117:e247–56.
6. Roberts RE, Roberts CR, Duong HT. Chronic insomnia and its negative consequences for health and functioning of adolescents: a 12-month prospective study. J Adolesc Health 2008;42:294–302.
7. Johnson EO, Roth T, Breslau N. The association of insomnia with anxiety disorders and depression: exploration of the direction of risk. J Psychiatr Res 2006;40:700–8.
8. Roberts RE, Duong HT. Depression and insomnia among adolescents: a prospective perspective. J Affect Disord 2013;148:66–71.
9. Bootzin RR, Stevens SJ. Adolescents, substance abuse, and the treatment of insomnia and daytime sleepiness. Clin Psychol Rev 2005;25:629–44.
10. Tynjala J, Kannas L, Levalahti E. Perceived tiredness among adolescents and its association with sleep habits and use of psychoactive substances. J Sleep Res 1997;6:189–98.
11. Dewald JF, Meijer AM, Oort FJ, Kerkhof GA, Bogels SM. The influence of sleep quality, sleep duration, and sleepiness on school performance in children and adolescents: a meta-analytic review. Sleep Med Rev 2010;14:179–89.
12. Meijer AM, Reitz E, Dekovic M, van den Wittenboer M, Stoel RD. Longitudinal relations between sleep quality, time in bed and adolescent problem behaviour. J Child Psychol Psychiatry 2010;51:1278–86.
13. Fischman S, Kuffler DP, Bloch C. Disordered sleep as a cause of attention deficit/hyperactivity disorder: recognition and management. Clin Pediatr 2015;54:713–22.
14. Norell-Clarke A, Nyander E, Jansson-Frojmark M. Sleepless in Sweden: a single subject study of effects of cognitive therapy for insomnia on three adolescents. Behav Cogn Psychother 2011;39:367–74.
15. De Bruin EJ, Oort FJ, Bögels SM, Meijer AM. Efficacy of internet and group-administered cognitive behavioral therapy for insomnia in adolescents: a pilot study. Behav Sleep Med 2014;12:235–54.
16. Schlarb AA, Liddle CC, Hautzinger M. JuSt - a multimodal program for treatment of insomnia in adolescents: a pilot study. Nat Sci Sleep 2011;3:13–20.
17. Clarke G, McGlinchey EL, Hein K, et al. Cognitive-behavioral treatment of insomnia and depression in adolescents: a pilot randomized trial. Behav Res Ther 2015;69:111–8.
18. De Bruin EJ, Bögels SM, Oort FJ, Meijer AM. Efficacy of cognitive behavioral therapy for insomnia in adolescents: a randomized controlled trial with Internet therapy, group therapy and a waiting list condition. Sleep 2015;38:1913–26.
19. De Bruin EJ, Bogels SM, Oort FJ, Meijer AM. Sleeping Smart improves adolescent psychopathology: results from a randomized controlled trial over 1 year. In: De Bruin EJ, ed. Insomnia Treatment for Adolescents: Effectiveness of group- and Internet therapy for sleep, psychopathology, cognitive functioning and societal costs. Delft: Uitgeverij Eburon, 2016:83–105.
20. Cheng TC. Factors related to adolescents' seeking help from social workers in mental health settings. Child Youth Serv Rev 2009;31:807–12.
21. Vitiello MV, McCurry SM, Rybarczyk BD. The future of cognitive behavioral therapy for insomnia: what important research remains to be done? J Clin Psychol 2013;69:1013–21.
22. Havas J, de Nooijer J, Crutzen R, Feron F. Adolescents' views about an internet platform for adolescents with mental health problems. Health Education 2011;111:164–76.
23. Nelson TD, Nelson JM. Evidence-based practice and the culture of adolescence. Prof Psychol Res Pr 2010;41:305–11.
24. Cushing CC, Steele RG. A meta-analytic review of eHealth interventions for pediatric health promoting and maintaining behaviors. J Pediatr Psychol 2010;35:937–49.
25. Stinson J, Wilson R, Gill N, Yamada J, Holt J. A systematic review of internet-based self-management interventions for youth with health conditions. J Pediatr Psychol 2009;34:495–510.
26. Briggs AH, Wonderling DE, Mooney CZ. Pulling cost-effectiveness analysis up by its bootstraps: a non-parametric approach to confidence interval estimation. Health Econ 1997;6:327–40.
27. Briggs A, Fenn P. Confidence intervals or surfaces? Uncertainty on the cost-effectiveness plane. Health Econ 1998;7:723–40.
28. Glick HA, Doshi JA, Sonnad SS, Polsky D. Economic evaluation in clinical trials. Oxford, UK: Oxford University Press, 2007.
29. Smulders YM, Thijs A. Kostprijs per gewonnen levensjaar: Trends en tegenstrijdigheden. Nederlands TijdschriftGeneeskunde 2006;150:2467–70. (The cost per year of life gained: trends and internal contradictions published in the Dutch Journal of Medicine).
30. Botteman M. Health economics of insomnia therapy: implications for policy. Sleep Med 2009;10:S22–5.
31. Daley M, Morin CM, LeBlanc M, Gregoire JP, Savard J. The economic burden of insomnia: direct and indirect costs for individuals with insomnia syndrome, insomnia symptoms, and good sleepers. Sleep 2009;32:55–64.
32. Fullerton DS. The economic impact of insomnia in managed care: a clearer picture emerges. Am J Manag Care 2006;12:S246–52.
33. Rosekind MR, Gregory KB. Insomnia risks and costs: health, safety, and quality of life. Am J Manag Care 2010;16:617–26.
34. Martin SA, Aikens JE, Chervin RD. Toward cost-effectiveness analysis in the diagnosis and treatment of insomnia. Sleep Med Rev 2004;8:63–72.
35. Bastien CH, Morin CM, Ouellet MC, Blais FC, Bouchard S. Cognitive-behavioral therapy for insomnia: comparison of individual therapy, group therapy, and telephone consultations. J Consult Clin Psychol 2004;72:653–9.
36. Scott GW, Scott HM, O'Keeffe KM, Gander PH. Insomnia - treatment pathways, costs and quality of life. Cost Eff Resour Alloc 2011;9:10.
37. Bonin EM, Beecham J, Swift N, Raikundalia S, Brown JS. Psycho-educational CBT-Insomnia workshops in the community. A cost-effectiveness analysis alongside a randomised controlled trial. Behav Res Ther 2014;55:40–7.

38. Bastien CH, Vallieres A, Morin CM. Validation of the Insomnia Severity Index as an outcome measure for insomnia research. Sleep Med 2001;2:297–307.
39. Watanabe N, Furukawa TA, Shimodera S, Katsuki F, Fujita H, Sasaki M, et al. Cost-effectiveness of cognitive behavioral therapy for insomnia comorbid with depression: analysis of a randomized controlled trial. Psychiatry Clin Neurosci 2015;69:335–43.
40. Tate DF, Finkelstein EA, Khavjou O, Gustafson A. Cost effectiveness of internet interventions: review and recommendations. Ann Behav Med 2009;38:40–5.
41. Arnberg FK, Linton SJ, Hultcrantz M, Heintz E, Jonsson U. Internet-delivered psychological treatments for mood and anxiety disorders: a systematic review of their efficacy, safety, and cost-effectiveness. PLoS One 2014;9:e98118.
42. Hedman E, Andersson E, Ljotsson B, Andersson G, Ruck C, Lindefors N. Cost-effectiveness of Internet-based cognitive behavior therapy vs. cognitive behavioral group therapy for social anxiety disorder: results from a randomized controlled trial. Behav Res Ther 2011;49:729–36.
43. Hedman E, El Alaoui S, Lindefors N, et al. Clinical effectiveness and cost-effectiveness of Internet- vs. group-based cognitive behavior therapy for social anxiety disorder: 4-year follow-up of a randomized trial. Behav Res Ther 2014;59:20–9.
44. Schulz KF, Altman DG, Moher D, Group C. CONSORT 2010 statement: updated guidelines for reporting parallel group randomised trials. BMJ 2010;340:c332.
45. Morin CM. Measuring outcomes in randomized clinical trials of insomnia treatments. Sleep Med Rev 2003;7:263–79.
46. Hakkaart-van Roijen L, Tan SS, Bouwmans CAM. Handleiding voor kostenonderzoek, methoden en standaard kostprijzen voor economische evaluaties in de gezondheidszorg. Geactualiseerde versie 2010 ed: College voor zorgverzekering (CVZ), 2010. [Dutch guidelines for cost research, methods and standard price units for economic evaluations in health care. Health Care Insurance Board (CVZ). Updated version 2010].
47. American Psychiatric Association. Diagnostic and statistical manual of mental disorders, 4th ed., text revision. Washington, DC: American Psychiatric Association, 2000.
48. Carney CE, Buysse DJ, Ancoli-Israel S, et al. The consensus sleep diary: standardizing prospective sleep self-monitoring. Sleep 2012;35:287–302.
49. EuroQol Group. EuroQol--a new facility for the measurement of health-related quality of life. Health Policy 1990;16:199–208.
50. Brooks R. EuroQol: the current state of play. Health Policy 1996;37:53–72.
51. Stolk EA, Busschbach JJ, Vogels T. Performance of the EuroQol in children with imperforate anus. Qual Life Res 2000;9:29–38.
52. Willems DCM, Joore MA, Nieman FHM, Severens JL, Wouters EFM, Hendriks JJE. Using EQ-5D in children with asthma, rheumatic disorders, diabetes, and speech/language and/or hearing disorders. Int J Technol Assess 2009;25:391–9.
53. Lamers LM, McDonnell J, Stalmeier PF, Krabbe PF, Busschbach JJ. The Dutch tariff: results and arguments for an effective design for national EQ-5D valuation studies. Health Econ 2006;15:1121–32.
54. Bodden DH, Dirksen CD, Bogels SM. Societal burden of clinically anxious youth referred for treatment: a cost-of-illness study. J Abnorm Child Psychol 2008;36:487–97.
55. Van Steensel FJ, Dirksen CD, Bogels SM. A cost of illness study of children with high-functioning autism spectrum disorders and comorbid anxiety disorders as compared to clinically anxious and typically developing children. J Autism Dev Disord 2013;43:2878–90.
56. Drogisterij.net. Drogisterij.net 2015. Accessed February 24, 2015. Available from: http://www.drogisterij.net.
57. Centraal Bureau voor de Statistiek. CBS StatLine 2015. Available from: http://statline.cbs.nl/Statweb/publication/?DM=SLNL&PA=71311ned&D1=0,4&D2=0&D3=123,142,155,168,181,194,219,232,245,258&HDR=G1,T&STB=G2&VW=T.
58. Acock AC. Working with missing values. J Marriage Fam 2005;67:1012–28.
59. Jacobson NS, Truax P. Clinical significance: a statistical approach to defining meaningful change in psychotherapy research. J Consult Clin Psych 1991;59:12–9.
60. Gradisar M, Crowley SJ. Delayed sleep phase disorder in youth. Curr Opin Psychiatry 2013;26:580–5.
61. Kerkhof GA, Geuke ME, Brouwer A, Rijsman RM, Schimsheimer RJ, Van Kasteel V. A new sleep disorders questionnaire based on the International Classification of Sleep Disorders-2. J Sleep Res 2012;22:104–7.
62. Morgenthaler T, Kramer M, Alessi C, et al. Practice parameters for the psychological and behavioral treatment of insomnia: an update. An American Academy of Sleep Medicine report. Sleep 2006;29:1415–9.
63. Morin CM, Bootzin RR, Buysse DJ, Edinger JD, Espie CA, Lichstein KL. Psychological and behavioral treatment of insomnia: update of the recent evidence (1998-2004). Sleep 2006;29:1398–414.
64. Schimsheimer RJ, Knuistingh Neven A, Jongh TOH. Slaapklachten. Huisarts en Wetenschap 2004;47:426–30.
65. Espie CA. "Stepped care": a health technology solution for delivering cognitive behavioral therapy as a first line insomnia treatment. Sleep 2009;32:1549–58.
66. Kyle SD, Espie CA, Morgan K. "…Not just a minor thing, it is something major, which stops you from functioning daily": quality of life and daytime functioning in insomnia. Behav Sleep Med 2010;8:123–40.
67. Lu M. The productivity of mental health care: an instrumental variable approach. J Ment Health Policy Econ 1999;2:59–71.
68. Little JDC. A proof for the Queuing Formula: $L = \lambda W$. Oper Res 1961;9:383–7.

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