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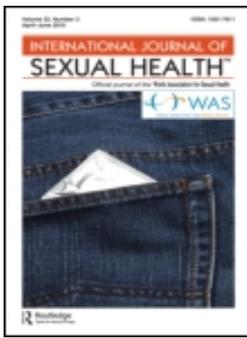
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ECONOMIC EVALUATION OF BRIEF SEX COUNSELING FOR SEXUALLY DYSFUNCTIONAL ADOLESCENTS

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ABSTRACT. *Objectives:* In a randomized controlled trial brief sex counselling (BSC), intensive sexual healthcare (ISH) and no treatment (NT) for adolescents with a sexual dysfunction were compared. The aim of this study was to assess the cost-effectiveness and cost-utility of BSC versus ISH and NT from a societal perspective. *Methods:* Costs, sexual functioning and quality of life were measured during 6 months. Primary outcome measures were measured with the Female Sexual Functional Index, the International Index of Erectile Function and the utilities reflecting Quality of Life based on the SF-36. Uncertainty was dealt with by using bootstrap replications and sensitivity analyses. *Results:* Results show that the societal costs were the highest for ISH followed by NT and BSC. The difference in costs between ISH compared to NT and BSC was significant. Furthermore, there were no significant group differences in sexual functioning or quality of life. With respect to the cost-effectiveness and cost-utility, BSC can be considered to be a suitable treatment for adolescents with a sexual dysfunction. *Conclusions:* Due to the lack of a significant difference in clinical effect, it can be concluded that BSC is the cheapest treatment option when implementing the intention-to-treat, besides a no treatment option, for adolescents with a sexual dysfunction.

KEYWORDS. Sexual dysfunction, adolescents and young adults, psychological treatment, cost-effectiveness

INTRODUCTION

Although Dutch adolescents are considered to be sexually healthy (Bakker & Vanwesenbeeck, 2006), Kedde (2012) reported that substantial numbers of adolescents in The Netherlands experience problems with sexual desire, sexual arousal, orgasm, and pain during sexual encounters. Female adolescents especially report these problems. Among girls, 43.4% experience at least one sexual dysfunction. Orgasmic disorders are the most frequently reported sexual dysfunction (20.3%). Among boys, 27.3% experience at least one sexual dysfunction. Premature ejaculation is the most frequently reported sexual dysfunction (12.6%). These findings can be taken to imply

that sexual function problems in adolescents warrant the attention of researchers, professionals, and policymakers.

When adolescents acknowledge that they experience a sexual problem, finding and receiving adequate care is often problematic. The loss of anonymity, financial costs of intensive sexual health care (ISH), and limited familiarity with and lack of confidence in the health care services available can be problems (Akre, Michaud, & Suris, 2010; Bakker et al., 2009; Bakker & Vanwesenbeeck, 2006). Goldmeier, Malik, and Green (2004) calculated that the average cost for treatment delivered by a sex therapist in the United Kingdom is approximately €900 (\$1,200). Given these costs, it is

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essential from a societal perspective that cost-effective treatment options are developed and evaluated. There are indications that this is possible for the treatment of sexual dysfunctions. For instance, 14% of the patients treated in outpatient clinics for sexology and 24% of the patients treated in mental health care centers could be adequately treated with a less costly, low-threshold intervention (Bakker & Vanwesenbeeck, 2006; Vroege, Lotgerink, van der Rhee, Tanis-Nauta, & Weijmar Schultz, 2007). However, the cost-effectiveness of these low-threshold interventions needs to be examined first to obtain the required financial support. There are published economic evaluations concerning HIV care showing that brief behavioral interventions to reduce the incidence of HIV and other sexually transmitted infections are cost-effective (Burgos et al., 2010; Zaric, Bayoumi, Brandeau, & Owens, 1998). However, to our knowledge, full economic evaluations concerning treatment options for adolescents who experience sexual dysfunctions are lacking.

In this study, we focus on “brief sex counseling.” Brief sex counseling (BSC) is a new treatment, developed by the Rotterdam-Rijnmond Public Health Service (PHS) and Maastricht University in The Netherlands, that could provide a more cost-effective treatment for adolescents with sexual dysfunctions.

In a randomized controlled trial, we examined the cost-effectiveness and the cost utility of BSC compared to ISH and no treatment (NT), while focusing on the following research question: Is BSC, compared to ISH and NT, preferable for the treatment of adolescents with sexual dysfunctions in terms of cost-effectiveness from a societal perspective?

METHODS

Participants

Participants were included if they met the following criteria: age 18 to 25 years old; at least mild to moderate sexual function problems; no severe psychiatric comorbidity or relational problems; and sexual function problems were not caused by disease, medication/drug use, or physical disabilities (based on a problem history

questionnaire). Participants were recruited through referrals from the PHS in Rotterdam-Rijnmond in the Northern parts of The Netherlands, and in Limburg (Southern part of The Netherlands), the Web site Sense, and various other Web sites (<http://www.sexcounseling.eu>, <http://www.sense.info> [The webpage is out of business now]). After random assignment, participants received their treatment in one of the following three regions in The Netherlands: Rotterdam, Northern Netherlands, and Limburg.

Procedure

This economic evaluation was embedded in a trial-based economic evaluation with a time horizon of 6 months. Details of the study are described in the design article (Grauvogel et al., 2009). After informed consent was obtained and participants completed an online screening questionnaire, they were randomly allocated to: (a) BSC, (b) ISH, or (c) NT. The outcomes and costs were measured via a secure Internet link (<http://www.emium.nl>) before random assignment (during initial screening), at baseline, and at 3 and 6 months after baseline as well as at 9 months after baseline only for ISH and BSC. To reduce the period that the respondents in the NT group were denied treatment, the last follow-up in this group was at 6 months after baseline. We used a block randomization procedure—and more specifically, an adaptive biased urn randomization for small strata—to ascertain that each intervention runs in equally large groups (Schouten, 1995).

Interventions

Brief sex counseling

BSC is a brief, free-of-charge, and low-threshold treatment for adolescents aged 18 to 25 years old who experience sexual function problems. In a maximum of three sessions, of 45 min each, over a 3-month period, a sex counselor provides treatment. The sex counselor is a nurse who works at Sense and who received supplementary training by a sexologist. The sessions include providing information

(psychoeducation) and cognitive restructuring. Instructions for home-based exercises, following the principles of the sensate-focus exercises of Masters and Johnson (1970), are also provided during the sessions.

The treatment manual that has been developed for the BSC sessions (Grauvogl & van Lankveld, 2009) is based on the general principles and practice of sex counseling and on a guideline that was developed for Sense (Ijff, 2006; Leusink & Nauta, 2009). The manual contains instructions for the sex counselor to address specific sexual problems. In the first session, specific questions regarding sexual functioning and overall sexual history are discussed. Furthermore, an action plan is established to provide an outline of the adolescent's treatment goals and the elements that will be used to alleviate the problem. Several therapeutic techniques are offered in Sessions 2 and 3. During the sessions, the counselor attempts to identify, change, and adjust compromised cognitive processes and deficient coping strategies. Instructions for sensate-focus exercises (Masters & Johnson, 1970) can be given to help the adolescent (re)discover sexual needs and preferences and to practice sensual and erotic touching, while avoiding performance pressure.

The brief duration of the intervention was expected to render BSC unsuitable to treat individuals with sexual dysfunctions with comorbid relationship and/or psychiatric problems.

Intensive sexual health care

In the ISH condition, participants received more intensive psychological treatment. During a minimum of six sessions during a 6-month period, a certified sexologist offers sexological treatment based on cognitive-behavioral therapy principles. The sessions are scheduled weekly. Furthermore, in contrast to BSC, the therapist in ISH also addresses the psychological problems of the participant, which may be the cause of the sexual function problem.

No treatment

The active treatment conditions were compared to the NT condition, which enabled the

researchers to control for spontaneous recovery and mere measurement effects. In this condition, participants were requested not to engage in other types of professional help for a 6-month period. After the waiting period, the participants who were allocated to this condition could choose their preferred treatment (BSC or ISH).

Cost Measures and Valuation

This economic evaluation was performed from a societal perspective, which implies that all relevant costs and outcomes were taken into account. As several countries have a more limited perspective, an additional analysis from a health care perspective was performed. Total costs were estimated using a bottom-up approach, where information on each element of service is multiplied by an appropriate unit cost and is summed to provide an overall total cost (Drummond, Sculpher, Torrance, O'Brien, & Stoddart, 2005). We assessed intervention costs, health care costs, patient and family costs, and costs in other sectors. For this study, we developed a cost questionnaire for this group based on existing questionnaires (Hakkaart-van Roijen, van Straten, Donker, & Tiemens, 2002; Koopmanschap et al., 2004), which identified all relevant cost aspects.

To measure the actual use of resources, data were obtained using combined sources (registrations by professionals and the cost questionnaire). Resources used relating to the interventions were based on the registered number of sessions that all professionals spent on the treatment. All use of resources by the adolescent in and outside the healthcare sector was measured by means of the cost questionnaire.

The valuation of health care costs and patient and family costs was based on the updated Dutch manual for cost analysis in health care research (Hakkaart-van Roijen, Tan, & Bouwmans, 2010). This manual recommends using standardized cost prices. Costs in other sectors, and especially productivity costs, were calculated by means of the friction cost method based on a mean added value of the Dutch working population. The friction cost method takes into account production losses confined

to the period needed (usually 90 days) to replace a sick employee. Absenteeism from school was calculated using the number of days the adolescents was absent from school. Cost prices are presented in 2012 euro values (Hakkaart-van Roijen et al., 2010).

Outcome Measures

Several instruments were used to assess the outcomes of the economic evaluation. The primary outcome measures were sexual functioning as measured in women using the Female Sexual Function Index (FSFI; Rosen et al., 2000) and sexual functioning as measured in men using the International Index of Erectile Function (IIEF; Rosen et al., 1997). High scores indicate healthy sexual functioning. To combine female and male sexual functioning data, Z scores were calculated within the female and male subgroups, resulting in average Z score means of 0, with 1 standard deviation for each participant.

Within the cost-utility analysis, utilities were derived using the Short Form-6D (SF-6D), which is a utility instrument based on the health-related quality-of-life questionnaire Medical Outcomes Study 36-Item Short Form Health Survey (SF-36; Ware & Sherbourn, 1992). The utility score is derived from 11 items on the SF-36 and is composed of six dimensions of health and was calculated using preferences elicited from a general population in the United Kingdom, the so-called Brazier algorithm (Brazier, Roberts, & Deverill, 2002). The utility values derived from the Brazier algorithm were used to compute the quality-adjusted life year (QALY) metric by using the area-under-the-curve method (Drummond et al., 2005). The results were a gain of 0.5 QALY because there was a measurement period of only 6 months.

Statistical Methods

All primary analyses were performed according to the intention-to-treat principle, including data from all participants regardless of whether they completed the measurements or not. Intermittent missing data were imputed by calculating the mean values of the previous and

subsequent time points. Missing data due to lost to follow-up (32%) were replaced by imputed values, using the last-observation-carried-forward technique. For the analyses, the Statistical Package for the Social Sciences Version 20.0 statistical software and Excel (for the bootstraps) were used.

To describe the cost data, arithmetic means were used. The incremental cost-effectiveness ratio (ICER; Prochaska & Velicer, 1997) was determined based on the incremental costs and effects of BSC compared with the ISH and NT. The ICER was stated in terms of incremental costs per outcome rate (increase in sexual functioning); the cost-utility ratio focused on the incremental cost per QALY gained.

Group differences in clinical effect (proportion of recovery) and quality of life were analyzed with nonparametric Kruskal-Wallis tests with a $p < .05$ significance level. Because costs were not normally distributed, we performed 5,000 bootstrap simulations.

The economic evaluation included a base-case cost-effectiveness and utility analysis and several sensitivity analyses. In the base case, the primary outcome measure for the cost-effectiveness analysis was change from the baseline score of sexual functioning measured by using standardized results of both the FSFI and IIEF at 6 months, and for the cost-utility analysis, the primary outcome measure was QALY at 6 months based on the SF-6D using the UK Brazier tariff. Nonparametric bootstrap simulations were used to quantify the uncertainty around the ICERs, yielding information about the joint distribution of cost and effect differences. To determine the cost utility, we used a willingness to pay per QALY of \$1,112 to \$12,236. This threshold was calculated with information provided by the Council for Public Health and Health Care [Raad voor de Volksgezondheid en Zorg] (2006).

Several sensitivity analyses were performed to test for the robustness of the findings. Except for the first sensitivity analysis (where only study completers were included), all analyses were executed using the base-case data set where missing data were imputed. All analyses used nonparametric bootstrap resampling techniques to explore the uncertainty around estimates of

cost-effectiveness and cost utility. First, participants who did not miss a single measurement were analyzed (ISH, $n = 17$; BSC, $n = 19$; NT, $n = 19$). Second, cost-effectiveness was calculated for male and female adolescents separately, because the results could be different when using the original questionnaires and not a standardized version. Third, because the physical components of the SF-36 might not be accurate for adolescents with a sexual dysfunction, cost utility was calculated using only the mental status-related components of the SF-36. Fourth, cost-effectiveness and cost utility were calculated from a health care perspective (i.e., only using the total health care costs in the analyses). Fifth, to account for any baseline differences, both cost-effectiveness and cost-utility analyses were calculated with a correction for baseline differences in costs. For this baseline correction, we used the delta adjustment method (van Asselt et al., 2009). Finally, cost-effectiveness was calculated using a longer time horizon of 9 months for the ISH and BSC intervention groups; for the NT group, these data were not available.

RESULTS

Baseline Characteristics

The recruitment population consisted of 580 adolescents, and 157 of them participated in the screening. Of these 157 participants, 44 participants were not eligible, mainly due to too high Center for Epidemiological Studies Depression Scale (Bouwma et al., 1995) and Brief Symptom Inventory scores (de Beurs & Zitman, 2005); 11 were not willing to participate; 30 did not respond to any e-mails; and 1 did not have complaints. Seventy-one adolescents were randomized to ISH ($N = 23$), BSC ($N = 27$), or NT ($N = 21$). The study sample consisted of 11 male ($M = 21.91$, $SD = 1.97$) and 60 female ($M = 21.72$, $SD = 1.85$) adolescents with a sexual dysfunction. Every participant that was randomly assigned in the study survived.

In Table 1, baseline characteristics of the three groups are described. There were no differences in demographic (age, gender, relationship status,

number of children, education, and employment status) or diagnostic (sexual functioning based on the FSFI and IIEF) baseline characteristics. Furthermore, there were no significant differences in costs between the three treatment groups at the start of the trial.

Costs

Table 2 presents the volumes of health care use during the 6-month follow-up period for each group. Furthermore, Table 3 presents the uncorrected costs of the different cost items. Costs were bootstrapped and stratified per group. Societal costs were the highest for ISH (\$2,752), followed by NT (\$822) and BSC (\$623). The difference between ISH on the one hand and NT and BSC on the other was significant, $U(2) = 28.17$, $p = .001$. Health care costs accounted for 78% of the total costs. Health care costs and patient and family costs were the highest for ISH (\$2400, \$117), followed by NT (\$344, \$53) and BSC (\$421, \$36). Costs in other sectors were the highest for NT (\$335), followed by ISH (\$234) and BSC (\$166). It can be concluded that the costs for the BSC group were the lowest in every sector.

Effects

Clinical effectiveness and QALYs

In the base-case analysis, the results show a mean sexual functioning of $-.024$ for ISH, $.12$ for BSC, and $.11$ for NT. There were no significant group differences, $F(1, 70) = 1.665$, $p = .20$. With respect to male and female adolescents separately, it was found that in male adolescents, both treatment groups (ISH and BSC) had improvement effect sizes that were not expected. Sexual functioning, as measured with the IIEF, was found to be deteriorated after treatment rather than improved. With respect to NT, sexual functioning—counter to expectations—improved. In female adolescents, both treatment groups yielded marginal effect sizes. Sexual functioning, as measured with the FSFI, was found to be improved after treatment. As

TABLE 1. Baseline Characteristics

	ISH (N = 23)		BSC (N = 27)		NT (N = 21)		<i>p</i>
Age, mean (years; <i>SD</i>)	22.30	(1.89)	21.41	(1.86)	21.38	(1.77)	.159 ^a
Female sex, <i>n</i> (%)	19	(82.6)	25	(86.2)	18	(85.7)	.909 ^b
Partner, <i>n</i> (%)	8	(34.8)	4	(13.8)	4	(19)	.636 ^b
No	14	(60.9)	22	(75.9)	16	(76.2)	
Yes, a man	1	(4.3)	3	(10.3)	1	(4.8)	
Yes, a woman							
Children, <i>n</i> (%)	22	(95.7)	28	(96.6)	21	(100)	.657 ^b
None	1	(4.3)	1	(3.4)			
One							
Education, <i>n</i> (%)	6	(26.1)	14	(48.3)	13	(61.9)	.471 ^b
Primary school	5	(21.7)	8	(27.6)	3	(14.3)	
Middle secondary education	8	(34.8)	4	(13.8)	3	(14.3)	
Higher secondary education	4	(17.4)	3	(10.3)	2	(9.5)	
Academic education							
Work, yes (%)	15	(65.2)	22	(75.9)	15	(71.4)	.583 ^b
BSI, mean (<i>SD</i>)	0.30	(0.30)	.29	(0.19)	0.27	(.25)	.727 ^c
CES-D, mean (<i>SD</i>)	8.09	(6.84)	5.81	(4.24)	4.7	(4.30)	.097 ^a
Sexual function, mean (<i>SD</i>) ^d	-0.24	(0.93)	0.14	(0.99)	0.09	(1.06)	.369 ^a
QSSD (Questionnaire for the Screening of Sexual Dysfunctions), mean (<i>SD</i>) Satisfaction	1.57	(2.57)	2.19	(2.09)	2.14	(1.80)	.201 ^c
Quality of life, mean (<i>SD</i>)	0.592	(0.052)	0.581	(0.059)	0.601	(0.039)	.187 ^a
SF-6D, UK Brazier tariff							
SEAR (Self-Esteem and Relationship Questionnaire), mean (<i>SD</i>)	0.22	(0.19)	0.40	(0.17)	0.34	(0.18)	.006 ^{*a}
CEQ (Credibility/Expectancy Questionnaire), mean (<i>SD</i>)	62.93	(16.25)	62.67	(17.76)			.864 ^a
Costs, mean, 95% CI	258.31 [96.5, 420.2]	102.60 [38.1, 167.1]	178.72 [-17.7, 375.1]				.371 ^c
Health care costs	25.93 [0.28, 51.6]	15.94 [-0.25, 32.1]	7.37 [2.8, 11.9]				.256 ^c
Patient and family costs	138.66 [-51.3, 328.6]	43.59 [8.7, 78.5]	158.70 [-140.4, 457.7]				.471 ^c
Productivity costs	422.92 [174.7, 671.1]	162.1 [69.6, 254.6]	344.75 [-14.3, 703.8]				.09 ^c
Societal costs							

^a*p* value calculated with analysis of variance *F* test.

^b*p* value calculated with Pearson chi-square test.

^c*p* value calculated with the Kruskal-Wallis test.

^d*z* score of the Female Sexual Functional Index and the International Index of Erectile Function.

*Significant at *p* < .05.

Note. ISH = intensive sexual healthcare; BSC = brief sex counseling; NT = no treatment; BSI = Brief Symptom Inventory (de Beurs & Zitman, 2005); CES-D = Center for Epidemiological Studies Depression scale (Bouwma et al., 1995).

expected, sexual functioning of female adolescents in the NT group further deteriorated.

Furthermore, the results show a mean QALY of .32 for ISH, .31 for BSC, and .31 for NT. There were no significant differences between the groups in QALYs, $F(1, 70) = .395$, $p = .68$.

Cost-Effectiveness and Cost Utility

The costs and level of sexual functioning of BSC versus ISH and BSC versus NT separately are plotted in cost-effectiveness planes (Figure 1a). BSC tends to be more effective and

has lower costs than ISH (90% of the bootstrapped pairs are lying in the cost-effective southeast quadrant). When looking at BSC versus NT, the majority of the replications (45%) also lie in the southeast quadrant, thus meaning that BSC is more effective and has lower costs than NT.

With respect to the costs and effects of cost per QALY (Figure 1b), the results show that BSC is more cost-effective than ISH (55% of the bootstrapped pairs are lying in the cost-effective southeast quadrant). With respect to the comparison between BSC and NT, the results show that the majority of the replications lie in the

TABLE 2. Volumes of Health Care Use During the 6 Months After Baseline

Health Care Items (Unit)	ISH			BSC			NT			Source Prices
	Mean (SD)	Max	Med	Mean (SD)	Max	Med	Mean (SD)	Max	Med	
Brief sex counseling (N sessions)	0.09 (0.29)	1	0	2.18 (1.24)	4	2	0.57 (0.98)	3	0	1
General practitioner (N consults)	2.33 (2.16)	6	2	2.15 (2.58)	10	1	2.25 (1.89)	6	2	2
Intensive sexual health care (N sessions)	7.65 (9.87)	48	6	0.11 (0.42)	2	0	0.00 (0.00)	0	—	2
Psychologist (N sessions)	0.39 (0.89)	3	0	0.11 (0.42)	2	0	0.15 (0.66)	3	0	2
Other mental health professional (N consults)	0.09 (0.29)	2	0	0.07 (0.27)	1	0	0.05 (0.22)	1	0	2
Medical specialist (N consults)	1.22 (2.75)	10	0	0.37 (0.93)	4	0	0.86 (1.46)	6	0	2
Company welfare worker (N consults)	0.27 (1.25)	6	0	0.11 (0.58)	3	0	0.00 (0.00)	0	—	3
Social worker (N sessions)	0.22 (1.05)	5	0	0.00 (0.00)	0	—	0.15 (0.65)	3	0	2
Center for Alcohol and Drugs (N consults)	0.04 (0.21)	1	0	0.11 (0.58)	3	0	0.00 (0.00)	0	—	1
Hospital day treatment (N half days)	0.22 (0.85)	4	0	0.00 (0.00)	0	0	0.00 (0.00)	3	0	2
Hospital stay (N days)	0.13 (0.63)	3	0	0.00 (0.00)	0	—	0.24 (0.89)	0	0	2
Self-help group (N sessions)	0.17 (0.38)	1	0	0.00 (0.00)	0	—	0.00 (0.00)	0	—	2
Prescribed medication, not for psychological disorders (€)	15.8 (40.8)	184	0	6.5 (16.5)	59	0	36.9 (84.9)	368	0	4
Over-the-counter medication (€)	3.4 (11.28)	47	0	7.0 (16.1)	50	0	8.9 (27.3)	117	0	4

Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment; a = including all forms of general practitioner consults (at the practice, at patient’s home, by telephone); 1 = cost prices according to public health service; 2 = cost prices according to Dutch standard cost prices of Hakkaart-van Roijen et al. (2010); 3 = cost prices according to other economic evaluation; 4 = cost prices according to <http://www.medicijnkosten.nl>.

southwest quadrant (47%), indicating that BSC has lower costs but is also less effective than NT. However, still, 39% of the replications lie in the dominant (southeast) quadrant.

Furthermore, the base-case cost-effectiveness analysis with the bootstrapped societal

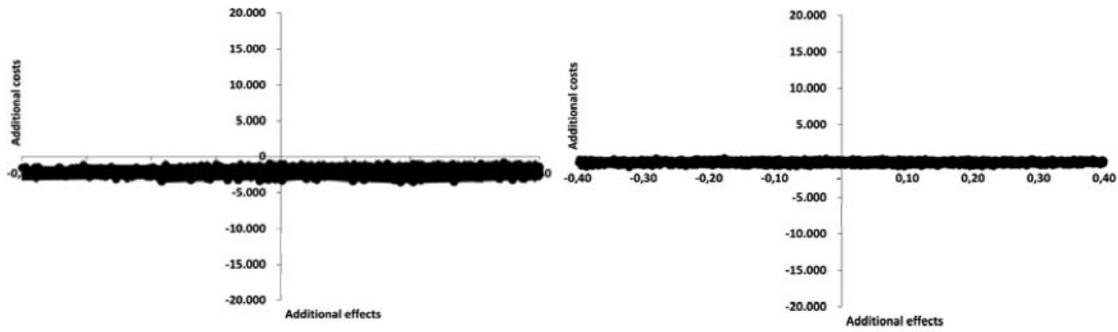
costs at 6 months and sexual functioning (standardized values of the FSFI and IIEF) as outcome is presented in Figure 2. BSC has a higher probability of being cost-effective than the other two comparisons (ISH and NT) up to a willingness to pay of \$39,206, varying from a

TABLE 3. Total Mean Costs in Euros Per Cost Item at 6 Months After Baseline (N = 71)

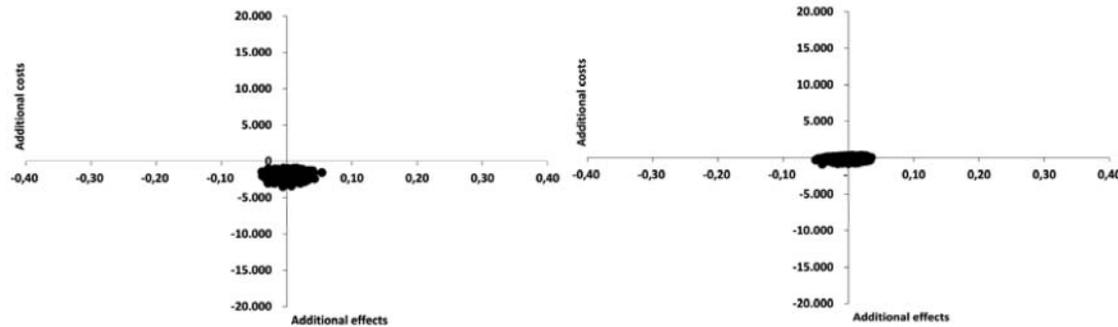
Cost Type	ISH (N = 23)	BSC (N = 27)	NT (N = 21)
Health care costs [95% CI]*	2,153 [1,508.0, 3,068.8]	378 [295.3, 478.8]	390 [203.9, 658.7]
Mental health care	1,727.05 (401.59)	265.71 (38.70)	70.96 (26.43)
Intervention	1,673.12 (403.84)	240.81 (117.57)	49.03 (18.18)
Other mental health care	53.93 (22.92)	24.90 (15.85)	21.93 (21.93)
General practitioner	81.45 (17.04)	61.87 (14.05)	65.73 (12.87)
Medical specialist	91.06 (42.43)	30.60 (14.65)	68.84 (25.37)
Company welfare worker	58.55 (16.75)	0.00 (0.00)	0.00 (0.00)
Social worker	13.15 (13.15)	0.00 (0.00)	10.36 (10.36)
Center for Alcohol and Drugs	3.29 (3.29)	8.29 (8.29)	0.00 (0.00)
Hospital day treatment	63.49 (51.73)	0.00 (0.00)	0.00 (0.00)
Hospital stay	69.36 (69.36)	0.00 (0.00)	121.38 (98.84)
Self-help group	1.90 (0.88)	0.00 (0.00)	0.00 (0.00)
Medication	42.06 (27.37)	12.95 (4.24)	52.93 (19.60)
Patient and family costs [95% CI]*	106.3 [71.2, 148.9]	32.7 [17.9, 53.8]	47.7 [17.1, 86.8]
Traveling	105.64 (19.66)	30.04 (8.84)	31.05 (13.52)
Informal care	0.63 (0.63)	2.66 (2.66)	16.60 (13.51)
Costs in other sectors [95% CI]*	210.0 [52.7, 479.1]	149.0 [64.4, 250.6]	300.8 [80.5, 649.6]
Productivity costs	70.83 (29.54)	106.47 (44.02)	183.97 (77.72)
Absenteeism from study	139.12 (101.08)	42.49 (21.02)	116.86 (85.70)
Societal costs [95% CI]*	2469.0 [1,750.8, 3,410.6]	559.7 [418.0, 720.5]	738.8 [396.6, 1,165.6]

Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

*The upper and lower confidence limits are the 2.5 and 97.5 percentile based on 5,000 bootstrap replications.



(a)



(b)

FIGURE 1. (a) BSC versus ISH—sexual functioning. BSC versus NT—sexual functioning. (b) BSC versus ISH—QALY. BSC versus NT—QALY. Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment; QALY = quality-adjusted life year.

probability of 79% at a cost-effectiveness threshold of \$0 to 34% at a cost-effectiveness threshold of \$36,238. When considering the threshold value of \$12,265, BSC has a probability of 42% of being the most optimal

treatment option, followed by NT with 38% and ISH with 20%. It should be noted that the difference between BSC and NT becomes indifferent after this point. The choice between the three study options

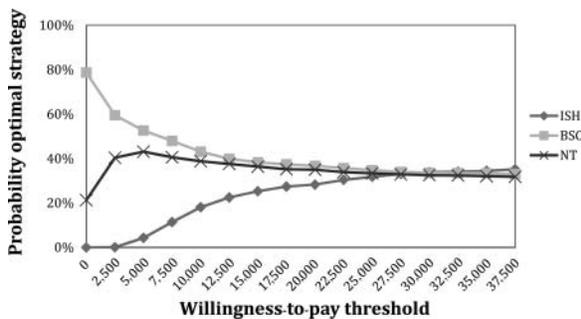


FIGURE 2. Cost-effectiveness acceptability curves looking at sexual functioning at 6 months after baseline. Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

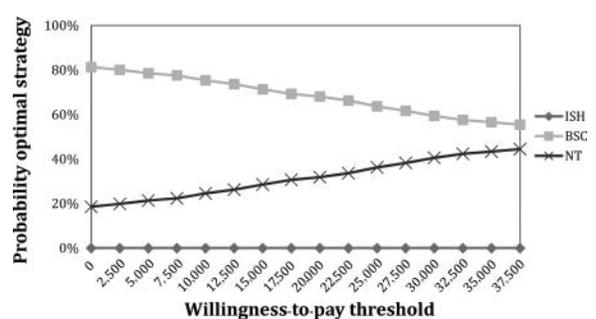


FIGURE 3. Cost-effectiveness acceptability curves looking at quality-adjusted life years at 6 months after baseline. Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

becomes indifferent, as all curves approach a probability of 33% when the threshold is higher than \$39,026.

The base-case cost-utility analysis with the bootstrapped societal costs at 6 months and the SF-6D as outcome is presented in Figure 3. With an 81% probability of being the most optimal treatment option at a threshold value of €0 per gained QALY and a probability of 75% at a threshold value of \$12,265 per gained QALY, BSC tends to be superior when compared with ISH and NT. The probability of being the most optimal treatment option increases for NT when the willingness to pay increases. However, up to a willingness to pay of \$55,272 per gained QALY, BSC has the highest probability to be the most optimal treatment option.

Sensitivity Analyses

1) Study completers only

In this scenario, data from study completers only were used. Results of the cost-effectiveness analysis (Appendix Figure 1) were comparable to the base-case analysis. Also here, BSC tended to be the most optimal treatment option when compared with ISH and NT; however, the difference between BSC and ISH is more prominent in favor of BSC. At any threshold, BSC has a higher probability of being cost-effective than the other two comparisons (ISH and NT). Overall, these results support the robustness of the base-case analysis. With respect to the cost-utility analysis (Appendix Figure 2), BSC has the highest probability of being the most optimal treatment option up to a willingness to pay of \$19,513.

2) Comparison of sexual functioning between male and female adolescents

In this scenario, sexual functioning was calculated for male (Appendix Figure 3) and female (Appendix Figure 4) adolescents separately. With respect to sexual functioning in female adolescents, the results are comparable with the base-case analysis. BSC is considered to be the best treatment option irrespective of

the willingness-to-pay threshold. With respect to sexual functioning in male adolescents, NT is considered to be superior over BSC and ISH. These results indicate that the results of the base-case analysis might be influenced by a gender effect.

3) Mental status-related components of the SF-36

In this scenario, only the mental properties of the SF-36 (Appendix Figure 5) were used because these aspects are thought to better reflect sexual problems. The results suggest BSC is the most optimal treatment option, up to a willingness to pay of \$19,513.

4) Analyses based on a health care perspective

In this scenario only health care costs were included in the analyses. Both cost-effectiveness (Appendix Figure 6) and cost utility (Appendix Figure 7) were comparable to the base-case analyses. However, the difference between BSC and NT is only very limited, suggesting that when only considering health care costs, BSC and NT are comparable treatment strategies.

5) Correction for baseline costs

In this scenario, the total costs were corrected for differences in baseline costs. Both cost-effectiveness (Appendix Figure 8) and cost utility (Appendix Figure 9) were comparable to the base-case analyses. The difference in cost-effectiveness between BSC and NT is only very limited, suggesting that when correcting for baseline costs, BSC and NT are comparable treatment strategies. However, the preference of BSC over NT is more prominent with respect to cost utility.

6) Cost-effectiveness at 9 months

In this scenario, a 9-month follow-up period (Appendix Figure 10) after baseline was used to compare ISH and BSC. The cost-effectiveness analysis was comparable to the base-

case analysis. BSC remains the most optimal treatment strategy. These results suggest that the effect remains stable even after a longer follow-up period.

DISCUSSION

The aim of the present study was to examine the cost-effectiveness and cost utility of BSC for adolescents with a sexual dysfunction. BSC was compared to ISH and NT. Based on both the cost-effectiveness and cost-utility analyses, BSC is a suitable treatment option for adolescents with a sexual dysfunction.

The results of the cost-effectiveness analysis indicate BSC as the most optimal treatment option, up to a willingness to pay of \$38,992. Although there were no significant differences in clinical effectiveness between the three groups, results show that in female adolescents, sexual functioning increases for BSC and ISH and decreases for NT. The initial increase in sexual functioning could be explained by the decrease, although not significant, in depressive symptoms and overall psychological problems. The lack of a significant difference in clinical effect indicates that this result is mainly caused by the difference in costs, not by effect.

In the cost-utility analysis, BSC was the most optimal treatment option. When closely looking at the division of costs, it can be concluded that societal costs are significantly higher for ISH than for BSC and NT. Mental health care costs, especially intervention costs, are relatively high in ISH. Furthermore, costs in other sectors were higher for NT, followed by ISH and BSC. The lack of guidance during the waiting period and the fact that their problems were not resolved might cause the higher absenteeism from school and work in this group.

Results of the sensitivity analyses, except for the results of the comparison between male and female adolescents, overall confirmed the robustness of the base-case analyses. The most important difference was the higher probability of BSC to be preferred over ISH and NT when only the data of study completers were analyzed. From a health care perspective and

when correcting for baseline costs, the difference in cost-effectiveness between BSC and NT was less prominent. Furthermore, results showed that when using the mental properties of the SF-36 only, the difference between BSC and ISH and NT was less conclusive than when the total SF-36 was used. An explanation for this finding could be that the SF-36 measures both physical aspects and mental aspects and that the mental aspects do not cover only psychological distress. This instrument might be less sensitive and responsive for adolescents with a sexual dysfunction (Wiebe, Guyatt, Weaver, Matijevic, & Sidwell, 2003). The best solution would be to develop a utility measure that also is suitable for people with sexual dysfunctions. The results of this study point out that a generic utility measure makes it more difficult to examine the cost utility for a specific mental disorder. Furthermore, this indicates that cost-utility analyses might be less suitable than cost-effectiveness analyses for adolescents with a sexual dysfunction due to the lack of a sexual component in the cost-utility analyses.

Methodological Considerations

Several methodological considerations concerning this study should be taken into account. First of all, this study had to deal with a substantial amount of missing values (32%). The drop-out of participants could be explained by two factors. First, due to the time between the measurements (3 months), it may have been difficult for the participants to remember what had happened during the previous 3 months and because of this they were not motivated to fill in the questionnaires. Furthermore, 65% of the participants dropped out after their treatment had finished. This finding could indicate that adolescents are motivated to complete questionnaires as long as they might be of interest for the treatment they receive. In adult populations, the dropout after treatment termination is not as high (Bamelis, Evers, Spinhoven, & Arntz, 2013; de Graaf et al., 2009). This suggests that when including adolescent participants in randomized controlled trials,

extra incentives should be provided in the follow-up period to ensure that they continue to complete questionnaires.

Second, there was a difference in the procedure of recording the data of the costs between the intervention costs and other costs. All costs were recorded using self-report data from the participants. With respect to intervention costs, we used the data received from the treatment centers yielding the actual number of sessions that the participant received. The number of sessions reported by the treatment centers did not always correspond to the number of treatment sessions reported by the adolescents. It might be possible that the self-reported costs were overestimated or underestimated, and as a consequence, they might have influenced the results.

Furthermore, there is no previous research that has examined the effect of a sexological treatment for adolescents with a sexual dysfunction, and consequently, there are no previous scientific data that provide information about the ceiling value of the willingness to pay. In previous research, various ceiling values per gained QALY have been mentioned and range from values of \$4,010 to \$164,047 (Counsel for Public Health and Health Care, 2006; Gyrd-Hansen, 2003; King, Tsevat, Lave, & Roberts, 2005). Thus, despite the absence of a clear guideline, it can be expected that the used ceiling value of up to \$12,254 is sufficient.

Conclusion and Recommendations

To our knowledge, this study is the first that has explored the cost-effectiveness and cost utility of sexological treatment of adolescents with a sexual dysfunction in general, and more specifically, the effect of BSC. The results of both the cost-effectiveness and cost-utility analyses suggest that BSC has the highest probability of being the most optimal treatment option. Despite the lack of a significant clinical difference between BSC on the one hand and ISH and NT on the other, based on the cost-effectiveness and cost utility, BSC might be considered to be a suitable treatment option for adolescents with a sexual dysfunction. However, it should be noted that

the cost-effectiveness is due to the difference in costs, not effect. Because the results of a single study are not sufficient, it is advised that researchers further investigate the effect of BSC. Furthermore, the effect of BSC might also be examined in adults with sexual dysfunction.

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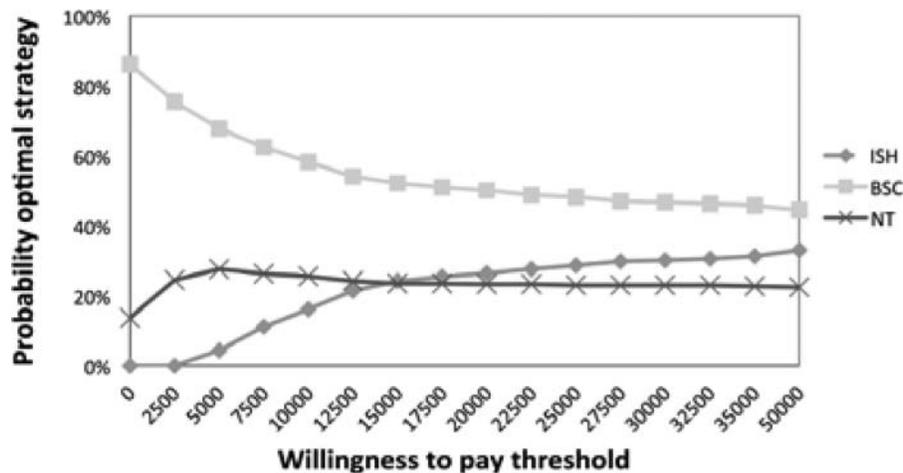
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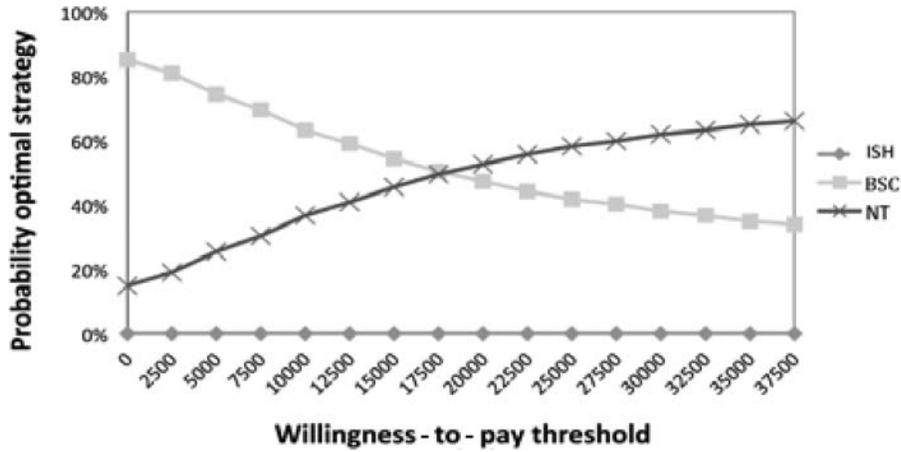
APPENDIX



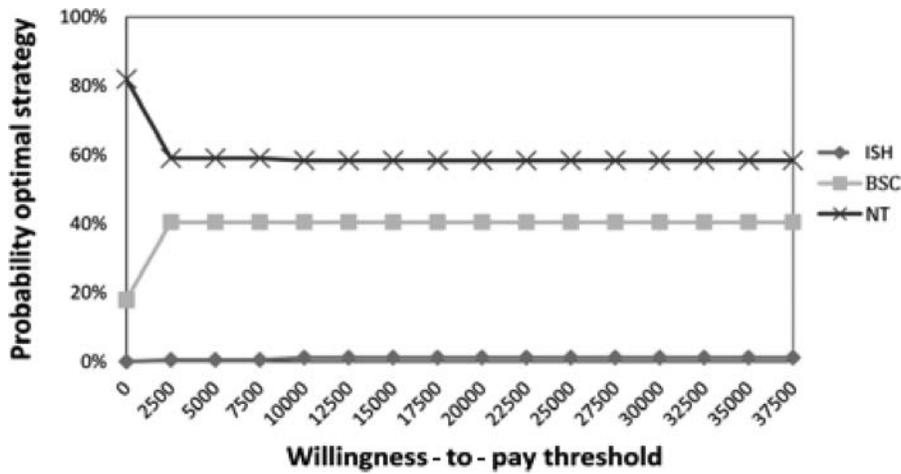
APPENDIX FIGURE 1. Cost-effectiveness, study completers.

Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

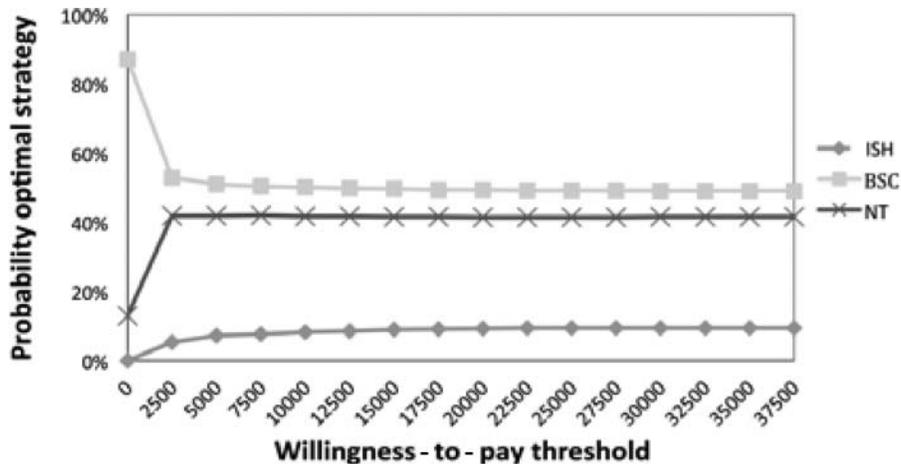
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APPENDIX FIGURE 2. Cost utility, study completers.
 Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

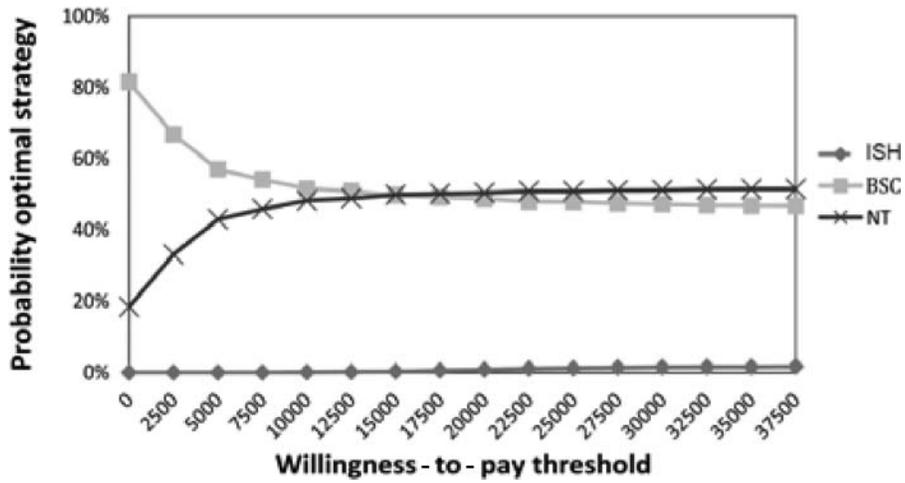


APPENDIX FIGURE 3. Cost-effectiveness, male adolescents.
 Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

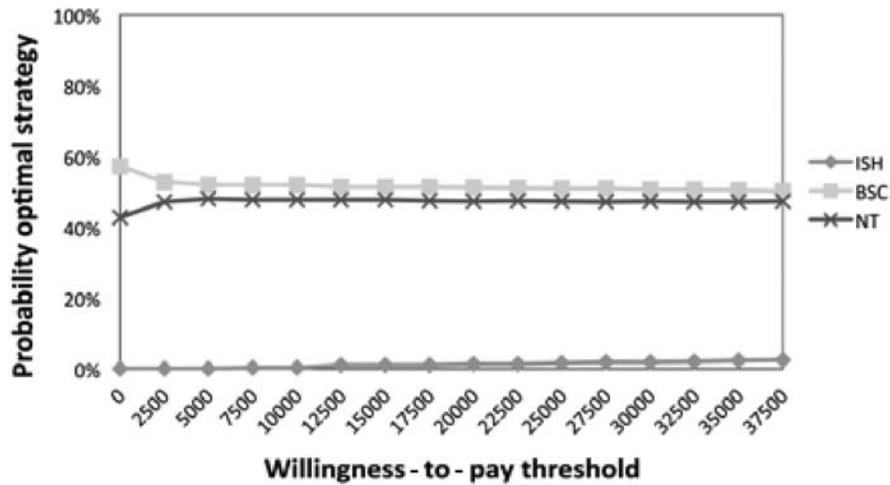


APPENDIX FIGURE 4. Cost-effectiveness, female adolescents.
 Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

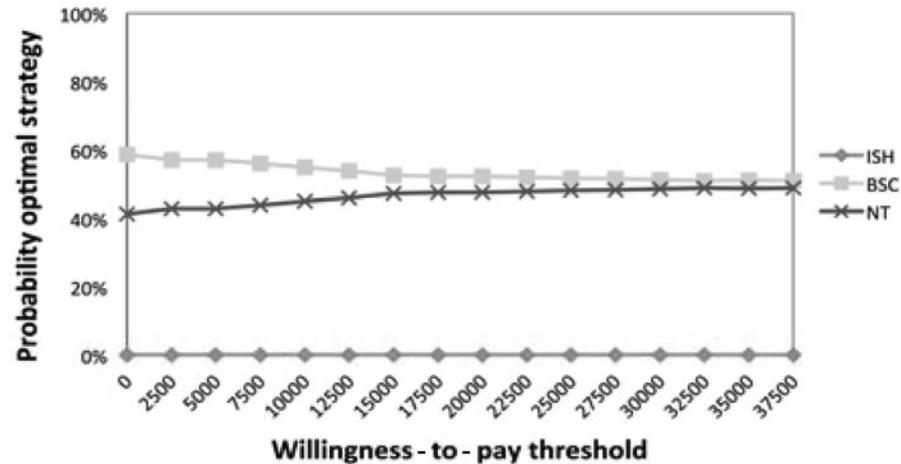
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APPENDIX FIGURE 5. SF-36 mental aspects.
 Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

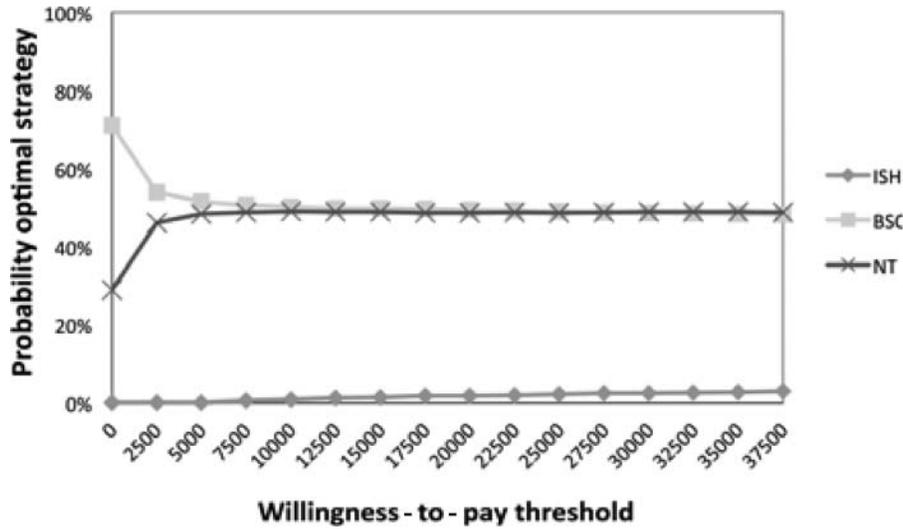


APPENDIX FIGURE 6. Cost-effectiveness, health care perspective.
 Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

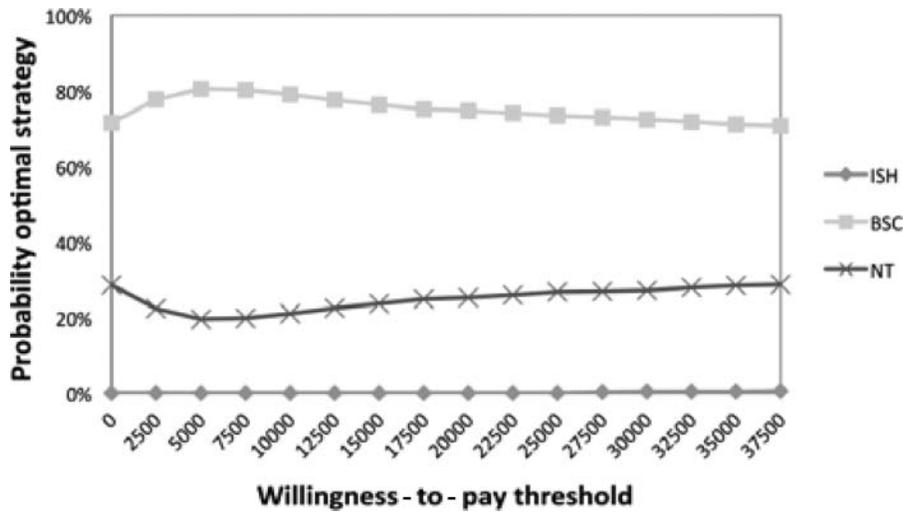


APPENDIX FIGURE 7. Cost utility, health care perspective.
 Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.

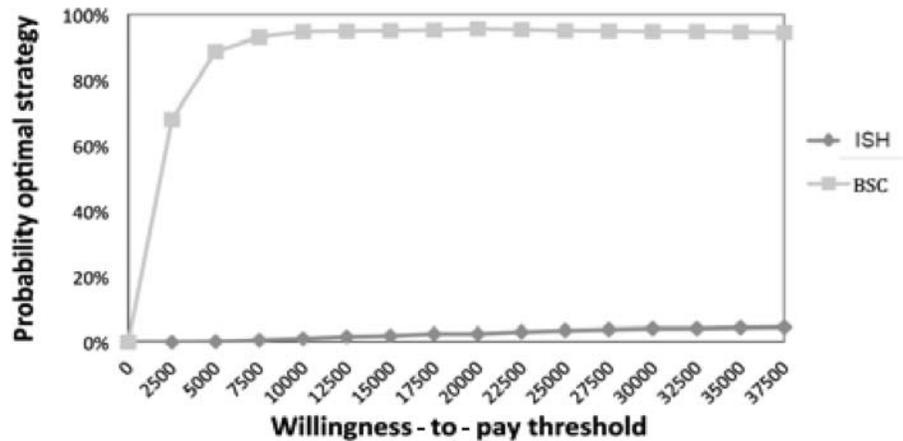
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APPENDIX FIGURE 8. Cost-effectiveness correction at baseline.
 Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.



APPENDIX FIGURE 9. Cost utility correction at baseline.
 Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.



APPENDIX FIGURE 10. Cost-effectiveness at 9-month follow-up.
 Note. ISH = intensive sexual health care; BSC = brief sex counseling; NT = no treatment.