

Willingness to Pay for Polysomnography in Children with Obstructive Sleep Apnea Syndrome: A Cost-benefit Analysis

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Objectives: To analyze willingness to pay (WTP) for polysomnography (PSG) among parents of children with obstructive sleep apnea syndrome (OSAS). To analyze the cost-benefit of PSG in a collectively funded healthcare system.

Setting: University-affiliated sleep laboratory.

Subjects: Parents of 158 boys and 94 girls, who had a mean age of 6.0 ± 3.9 years. The telephone survey, using a contingent valuation approach, was conducted with 3 groups of parents: those whose children were scheduled for PSG (n = 83), whose children were had had PSG within the previous 6 months (n = 77), and whose children had had PSG and adenotonsillectomy in the previous 6 months (n = 92).

Results: Two hundred and fifty-two parents (92% compliance rate), 75% of whom were mothers, responded to the WTP interview. Multivariate analysis revealed that the independent variables influencing WTP were bid (OR = 0.745, *P* < .001), age times bid (OR = 0.835, *P* < .05), and

affected health status (OR = 3.5, *P* < .001). The median WTP value for PSG studies of children with OSAS following adenotonsillectomy was \$762 plus the savings of \$60 to the health care system—subtracting the cost of the \$250 PSG study resulted in a monetary benefit of \$572 per diagnosis.

Conclusions: We conclude that PSG diagnosis for children with OSAS is beneficial. Decision makers and sleep specialists can use WTP to prioritize allocation of resources to increase the availability of PSG studies for children.

Key Words: Children, cost-benefit analysis, obstructive sleep apnea syndrome, polysomnography, willingness to pay.

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INTRODUCTION

OBSTRUCTIVE SLEEP APNEA SYNDROME (OSAS) IN CHILDREN IS A COMMON CHRONIC CONDITION, AFFECTING 2% TO 4% OF ALL CHILDREN, AND MAY BE ASSOCIATED WITH RECURRENT RESPIRATORY TRACT INFECTIONS, GROWTH FAILURE, COR PULMONALE, SECONDARY ENURESIS, BEHAVIORAL AND NEUROCOGNITIVE PROBLEMS, GROWTH RETARDATION, AND POOR SCHOOL ACHIEVEMENT.¹⁻⁶ The most common cause of OSAS in children is adenotonsillar hypertrophy, thus adenotonsillectomy is the treatment of choice.⁷ Following adenotonsillectomy, most children with OSAS will have both symptomatic and polysomnographic (PSG) resolution of the disorder, as well as a significant improvement at least in short-term quality of life.⁸

Obstructive sleep apnea syndrome has attracted the attention of healthcare policymakers in insurance plans and health maintenance organizations because the volume of referrals for PSG studies is steadily increasing. Children with OSAS are heavy consumers of healthcare resources; adenotonsillectomy has been found to decrease healthcare utilization 1 year following the operation.^{9,10} Decision makers in the healthcare markets are concerned with efficient allocation of scarce resources, in particular, with making choices about prioritizing different treatments, including PSG evaluation. Cost-benefit analysis allows decision makers to rank the benefits per unit of a given budget and to use this

ranking to allocate the limited budget among the competing requirements. Cost-benefit analysis is an alternative approach to cost-effectiveness analysis for evaluating healthcare interventions.¹¹ The primary difference between cost-effectiveness analysis and cost-benefit analysis is the way in which health benefits or outcomes are measured. Cost-benefit analysis measures both costs and health outcomes in a common money metric,¹²⁻¹⁵ while cost-effectiveness analysis measures health benefits in nonmonetary units such as life-years gained.¹¹

In order to evaluate the benefits of a health intervention, it is common to measure the public's willingness to pay (WTP) as a part of cost-benefit analysis. The concept of WTP was developed primarily to value environmental benefits.¹⁶ Recently, WTP has been used to value health services.¹⁷⁻²⁰ Willingness to pay measures the monetary evaluation of a service to its consumer; WTP for health treatment is the maximum amount of money an individual who suffers from an illness would pay for a treatment that restores his or her health. To estimate aggregate patient demand for a medical treatment, it is assumed that each patient would purchase the medical treatment if the price were less than or equal to his or her WTP.¹⁶ In areas of public-sector activity such as health care, where conventional markets do not exist, decisions still have to be made about how best to use limited resources. This requires valuation of both resource costs and benefits of interventions, ie, resources saved, health gained, and other sources of well being. For goods traded in the private markets, the WTP can be directly observed from consumer-purchasing behavior. If the healthcare intervention were sold in the private market, it would not be necessary to conduct WTP surveys because the public's preferences would be revealed in the market. With this type of information, a combination of interventions can be chosen to maximize the value of benefits to the community.¹¹

Polysomnographic study is the gold-standard diagnostic approach for OSAS. It has been shown to be cost effective compared with alternative diagnostic approaches.²¹ The results of PSG studies enable physicians as well as parents to decide on the best treatment alternatives. In pediatric OSAS, otolaryngologists may prefer to perform an adenotonsillectomy

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No significant financial interest/other relationship to disclose.

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based solely on the presence of clear clinical symptoms. The present paper focuses on parents' WTP for a child's PSG in which the child's presenting symptoms without a PSG study did not clearly indicate the need to perform an adenotonsillectomy. This economic methodology offers new insight on the relative importance of a specific aspect of diagnosis in OSAS. We interviewed 3 groups of parents during different stages of OSAS diagnosis and treatment. These groups represent the parents of a mixture of patients who are suspected of having or who have been diagnosed with OSAS, parents who are involved in the diagnostic and therapeutic processes.

METHODS

The data for this cross-sectional study were collected between September and December 2001 at the Pediatric Sleep Disorder Center in the Soroka University Medical Center, the only center in the southern region of Israel. The Center is affiliated with the Faculty of Health Sciences of Ben-Gurion University of the Negev, Israel. According to the Israeli National Health Insurance Law,²² patients should receive a PSG study and all diagnostic and treatment information regarding OSAS free of charge. Clalit Health Care Services has adopted a free-access policy to PSG studies for children suspected of having OSAS. Physicians are paid a capitation fee once every 3 months per patient and, therefore, do not have any economic incentive to increase consumption of services or to prevent a PSG study from taking place. No barrier, including copayment, exists for children referred for a PSG study. Therefore, parents included in the study represent all socioeconomic levels of the population in our region.

Subjects

The study subjects included 3 groups of consecutively recruited parents (mother or father) of 273 children during the different diagnostic and therapeutic stages of OSAS; Group 1 comprised parents of children scheduled for PSG study; Group 2, parents of children 1 week to 6 months following the PSG study; and Group 3, parents of children following PSG and adenotonsillectomy. All families had been permanent residents of the region for at least 3 years prior to study initiation and are enrollees of Clalit Health Care Services, the largest health maintenance organization in Israel. All subjects participating in the study were from the Negev, the southern region of Israel. This region includes approximately 145,000 children, of whom 60% are Clalit Health Care Services members. We excluded parents of children under the age of 1 year and children with additional chronic morbidity.

The project was approved by the Ethics Committee of Ben-Gurion University of the Negev.

Data Collection: Telephone Questionnaires

Sleep Questionnaire

The diagnostic evaluation included a detailed clinical history obtained using a standard questionnaire.²³

Willingness to Pay Questionnaire

Appendix 1 includes the 3 versions of the WTP questionnaire adapted to the 3 groups investigated. We used the contingent valuation approach, which is currently accepted in the healthcare market as a method to evaluate WTP for goods not traded in the private markets.^{17,24} One technique in the contingent valuation approach is the binary contingent valuation questionnaire using a bidding game in which respondents are asked whether they are willing to pay an offered bid for a specific good, assuming the good is available in the private market. The WTP estimation obtained is interpreted as the amount of income the patient is willing to give up to improve his or her health level. The bid amount varies across respondents.²⁵⁻²⁸ Median WTP is calculated as the public value emanating from the use of the PSG study in monetary terms. The amounts used

for the bid ranged from 25% to 10 times more than the actual price of PSG studies in Israel during December 2001, ie, 250, 450, 850, 1050, 1250, 2000, 2500, 4000, 6000, and 10,000 new Israeli shekels. The highest and lowest bid values were included to ensure that the upper and lower limits were reached. The bid results were converted from new Israeli shekels to United States dollars (4.2 new Israeli shekels = \$1).

Socioeconomic level was evaluated indirectly according to residential location (mailing address) as defined and monitored by the Central Bureau of Statistics of Israel.²⁹ Each residential location was given a numeric value that, in turn, was clustered into 20 different socioeconomic levels. Level 1 reflects a population with the lowest socioeconomic level in the country, and level 20 reflects a population with the highest socioeconomic level in the country.³⁰ The median socioeconomic level of subjects included in each of the bid groups was 5 (range, 4-19), and there is statistical differences found between the groups.

Polysomnographic Study

Overnight PSG study and scoring procedures were reported previously by our laboratory.¹⁰ Briefly, all children in groups 2 and 3 underwent nocturnal PSG monitoring (SensorMedics Inc., Yorba Linda, Calif, USA). Subjects reported to the sleep laboratory at 8:30 PM, were discharged at 7:30 the following morning, and were encouraged to maintain their customary daily routine and take medications as usual. Sleep and wake stages, arousals, and awakenings were scored as recommended with the appropriate modifications for children. The arousal index (AI) was calculated as the number of arousals or awakenings per hour of sleep. Obstructive apnea was defined as paradoxical breathing for at least 2 respiratory cycles with complete cessation of nasal airflow. Obstructive hypopnea was scored when the paradoxical breathing was accompanied by a reduction of at least 50% in airflow, resulting in either an arousal or an oxygen desaturation of at least 4%. The respiratory disturbance index (RDI) was defined as the number of apneas and hypopneas per hour of sleep. The average waiting time for a PSG study was approximately 7 weeks. Following a PSG diagnosis, treatment alternatives included adenotonsillectomy and follow-up visits.

Research Protocol

We conducted a telephone survey lasting 10 to 15 minutes to complete the WTP questionnaire. Parents were interviewed by 1 of our investigators (TS). During the interview process, the investigator gave explanations as needed. All interviews were conducted in Hebrew. In about 20 of the 252 cases where further clarification of the language was required, we interviewed a family member or neighbor with a good understanding of Hebrew. In addition, we used simple terms in order to simplify communication.

There were 10 groups of parents, randomly grouped using a random number table according to the bid values to be employed; 22 to 28 respondents were included in each bid group. A sample size of 252 parents revealed a statistical power above 0.8. Each bid was represented by an equal number of subjects from each of the 3 study groups. For patients on the PSG schedule, the interview was conducted no more than 8 weeks prior to the PSG study. For patients who had already completed the PSG study, the interview was conducted no more than 6 months following the PSG study. For patients having an adenotonsillectomy, the interview was conducted 4 weeks to 4 months after the operation.

Data Analysis

The data were collected into a Microsoft Access database (Microsoft Corp, Redmond, Wash, USA). Parametric tests (1-way analysis of variance [ANOVA]) was used when appropriate (for normally distributed variables) such as age, RDI, and AI. For nonnormally distributed variables, we used Kruskal-Wallis, ANOVA, median test. Data analysis was performed using SPSS for Windows (SPSS, Inc., Chicago, Ill, USA) and EViews (Quantitative Micro Software, Irvine, Calif, USA). We used

logistic regression to estimate WTP¹⁷ in univariate and adjusted WTP by using multivariate analyses with the following objective independent variables: age, bid, socioeconomic level, RDI, AI, and did or did not undergo adenotonsillectomy.

For the purpose of the study, we defined subjective variables *affected* and *nonaffected health status* for each of the 3 groups (denoted health status). For group 1 (child scheduled for PSG study) nonaffected health status was defined as, *I'm not or little worried* and affected health status was defined as, *I'm worried, very worried, and I think that my child's life is in danger*. For group 2 (child who underwent PSG study), nonaffected health status was defined as, *I have felt no or little relief*, and affected health status was defined as, *I felt some, significant and highly significant relief*. For group 3 (child who underwent PSG and adenotonsillectomy), nonaffected health status was defined as, *There was no or little improvement*, and affected health status was defined as *There was some, significant, or highly significant improvement*.

Savings to the healthcare system was calculated as the difference in total annual costs for healthcare utilization before and after adenotonsillectomy intervention.⁹

According to Drummond et al,¹¹ cost benefit in monetary values is equal to the adjusted WTP plus savings to the healthcare system times the proportion of children who underwent adenotonsillectomy with the cost of the PSG study then subtracted from that number. Recent findings from our laboratory⁹ demonstrated that the amount of savings to the healthcare system 1 year following adenotonsillectomy in children with OSAS was 34% (\$102). The proportion of children with OSAS who underwent adenotonsillectomy was 0.59. The actual cost of a PSG study as determined by the Israeli Ministry of Health is \$250.

RESULTS

Study Population

Two hundred and fifty-two parents (Table 1) agreed to participate in the telephone survey (compliance rate of 92%). Of the all respondents, 75% were mothers. The 158 boys and 94 girls had a mean age of 6.0 ± 3.9 years. Significant differences were found in group 3 compared with groups 1 and 2 with respect to age of the child and socioeconomic level. Twenty-six percent of the population had 2 children per family. Forty-nine percent of the children did not share their bedroom, 41% shared the room with 1 sibling, 8% with 2 siblings, and 2% with 3 or more siblings. The RDI and AI presented in Table 1 for group 3 were obtained from the PSG studies conducted before the children underwent the adenotonsillectomy. Children in groups 2 and 3 had RDIs compatible with moderate OSAS. The OSAS was more severe among the children in group 3 ($P = .01$), who showed significant sleep disruption as indicated by the AI ($P = .01$).

Thirty-two percent of the parents in group 1 reported they were only a little or not worried at all about their child's health level, while 68% of the parents in the same group reported themselves to be worried to extremely worried. In group 2, 41% felt little or no significant relief following the PSG study, while 59% of the parents in the same group reported some to significant relief following PSG diagnosis. Nine percent of the parents in group 3 reported little or no significant improvement in health level following the adenotonsillectomy, while 91% of the parents reported some to significant improvement following adenotonsillectomy.

Evaluation of WTP Bids

Univariate analysis revealed that socioeconomic level, RDI, and AI did not affect WTP for the PSG study. Figure 1 illustrates the percentage of parents' WTP in relation to the amount of the bid. The nonadjusted median WTP as a function of the bid was \$705. The higher the bid, the fewer parents there were WTP for PSG studies ($P = .0007$). Willingness to pay is affected as a function of age (age x bid): Age difference per se affected the parents' WTP. Thus, comparing 2 groups of parents having children with age differences of more than 12 years, parents of older children were less inclined to pay any bid compared to parents of younger children (odds ratio [OR] = 0.697, $P = .04$). When comparing 2 groups of parents having children with age differences of 2 years, parents of older children were inclined to pay and bid almost the same as parents for younger children (OR = 0.94, $P = .04$). Univariate logistic regression analysis revealed that parents of patients who underwent adenotonsillectomy were 1.8 times more WTP for a PSG study, compared with parents whose child did not undergo adenotonsillectomy (OR = 1.77, $P = .06$). Parents of patients who did not undergo adenotonsillectomy expressed a median WTP of about 2.8 times more than the actual market price.

Table 2 presents multivariate analysis determining the independent variables influencing WTP. Average WTP was found to be \$762 and was

Table 1—Patient Characteristics*

	Waiting for PSG n, 83	After PSG n, 77	After AT† n, 92
Boys/girls, no.	52/31	48/29	58/34
Age ≥8 y, %	35	36	17
Age, y	6.4 ± 4.0	6.9 ± 4.4	5.0 ± 3.2‡
Median socioeconomic level (range)§	10 (4-19)	10 (4-19)	10 (4-19)¶
RDI, events/h	-	6.8 ± 10.4	9.7 ± 9.0‡
AI, events/h	-	19.6 ± 10.1	25.2 ± 14.4‡

PSG refers to polysomnography; AT, adenotonsillectomy; RDI, respiratory disturbance index, the number of apneas and hypopneas per hour of sleep; AI, arousal index, number of arousal events per hour of sleep.

*Values are mean ± SD except as otherwise noted and were compared using 1-way analysis of variance except as otherwise noted.

†Children with obstructive sleep apnea syndrome who underwent AT. The RDI and AI presented in this group were obtained from the PSG study prior to AT.

‡ $P < .01$

§Socioeconomic level was evaluated indirectly according to residential location as defined by the Central Bureau of Statistics of Israel. Socioeconomic level 1 reflects a population with the lowest socioeconomic level, and 20 reflects the highest socioeconomic level. Values were compared with the Mann-Whitney test.

¶ $P < .02$

Table 2—Multivariate analysis: independent variables influencing willingness to pay

Variable	Coefficient	OR	95% CI	P value
C	1.828	0	-	-
Bid	-0.00124	0.745*	0.745-0.746	.0007
Age-Bid	-0.00012	0.835†	0.835-0.835	.046
Health status	-1.25	3.49‡	2.8-4.1	.0002

OR refers to odds ratio; CI, confidence interval

*Odds ratio obtained for a parent of a child 6 years of age and a bid difference of \$150 [= exp 150 (-0.00124-0.00012 x 6)].

†Odds ratio obtained for a bid of \$250 and age difference of 6 years [= exp (250 x 6 x -0.00012)].

‡Odds ratio obtained as exp 1.25. The coefficients of this logistic regression are used for calculating the average willingness to pay [= (-1.828+1.25 x 0.267)/(-0.00124-0.00012 x 6)], where the average health status is 0.267 and the average age is 6.03 years.

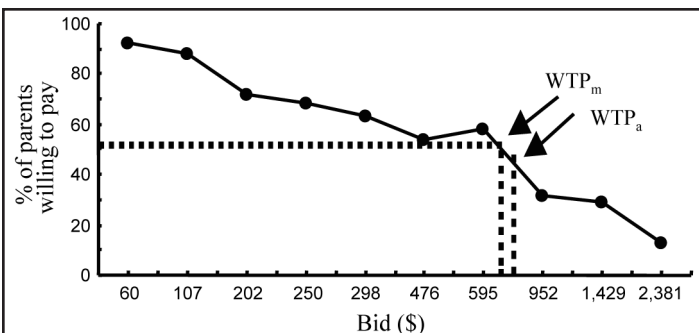


Figure 1—The relationship between bid amount (US\$) and willingness to pay (WTP). The higher the bid, the fewer parents willing to pay ($\beta = -0.000294$, $P = .0007$). WTP_m refers to median willingness to pay as a function of bid amount; WTP_a, adjusted WTP; WTP_m = \$705; WTP_a = \$762.

influenced by bid, function of age (age x bid), and health status. Since WTP is influenced by the bid, we analyzed the sensitivity of bid differences of \$100 or \$150, selected arbitrarily. For example, ORs for the bid imply that a payment decrease of \$150 (for a parent of a 6-year-old child) will increase the probability for WTP acceptance by 34% (OR = 1.34, $P = .0007$). As age rises, the OR for WTP decreases, ie, for 6 years of age difference, the OR for WTP is 0.835 ($P = .046$) and for a 16-year-old adolescent, the OR is 0.62 ($P < .05$). The most influential variable that affected WTP was health status (OR = 3.5, $P < .001$). This implies that for parents who felt relieved following diagnosis, the probability of accepting any payment (at any age) was 3.5 times greater than for parents who did not report any change in health status. Among the parents who did not report a change in their child's health status, WTP was about \$290, close to the actual market price of \$250 for the PSG study.

Cost-benefit Analysis

The PSG study results in a benefit of \$572 per patient. The benefit is equal to \$762 (the adjusted WTP) plus \$60.30 (savings to the healthcare system⁹) minus \$250 (the cost of PSG study) for a sum benefit of \$572. The benefit-to-cost ratio is 3.16, namely, benefits per patient are about 3 times higher than the cost.

DISCUSSION

In the present study, we measured WTP in a collectively funded healthcare program. Cost-benefit analysis revealed that a PSG study for children suspected of having OSAS is beneficial. The WTP is affected by the bid, age times the bid, and health status. Incorporation of subjective variable health status in the model showed that parents who felt relief or worry were 3.5 times more WTP for a PSG study than those who felt no relief or no worry about their child's health status. Parents of younger children are more WTP for a PSG study compared with parents of older children.

Study Population

The present study demonstrates results for *typical* parents of otherwise healthy children presenting clinical symptoms of OSAS. These typical parents may be regarded as representing parents of children in our region with a potential diagnosis of OSAS, since our sleep laboratory provides service to all enrollees in our referral area. The WTP may not represent all patient populations nationally and internationally. The WTP should be determined for different regions, ie, respondents in the United States are more likely than respondents in much of Europe and Israel to consider wage losses because of the absence of a comprehensive income security program in the United States.³¹ Thus, caution should be taken when comparing the WTP in different areas.

The WTP scenario as posed by the interviewer may affect the maximal WTP.^{31,32} We used the contingent valuation approach, a widely accepted technique^{17,28,31-35} for estimating WTP for goods not traded in the private market and to value treatment or progress in health.^{20,31,33} Since PSG studies are provided at no charge, we explained to the parents that they had to consider WTP according to a scenario in which they were required to prioritize out-of-pocket money to purchase the PSG study, the cost of time spent performing the PSG study, the expected health benefits, and the changes in quality of the child's life.³¹ During the interview, we did not provide information regarding aspects of the diagnostic process or external or spillover benefits to the family members. However, this latter information was provided to all patients as part of the routine in the pediatric sleep clinic.

A clear and tangible scenario is a key element in analyzing WTP, and studies usually must be carried out face to face with the interviewee.³² However, we conducted a successful telephone survey with a compliance rate of 92%. This was due to the clear messages given to parents regarding the importance of PSG study, followed by a simple and understandable scenario presented by 1 of our investigators (TS) during the

telephone survey. Other WTP studies have reported compliance rates between 65% and 75%.¹⁶⁻¹⁸ Further studies are needed to confirm our results in face-to-face WTP interviews. In our view, the scenario presented to parents was reasonable. The WTP bids given by the parents were within a reasonable and realistic range. The monetary value represented by the parents' WTP for a PSG study was 3 times more than its actual market cost and about half of the average monthly wage level in Israel.²⁹

Scale Effects on WTP for PSG Studies

This study presents a novel concept of cost-benefit analysis not yet studied in sleep medicine, specifically the WTP for PSG studies in children with a suspected diagnosis of OSAS. Therefore, we selected arbitrary independent variables that may affect WTP and provided evidence that this concept is viable. Health status was chosen because clinical assessment by physicians has a pure diagnostic value for OSAS.¹ Therefore, it was of interest to find out how parents perceive the importance of the PSG study in determining the final diagnosis. Other variables such as age, socioeconomic level, and RDI are, in our experience,¹⁰ outcome measures that predict morbidity and healthcare utilization associated with OSAS. We acknowledge the possibility that other important variables, such as the child's place in the family and previous siblings with OSAS, and outcomes, such as associated morbidity, quality of life, and school and neurocognitive performance, may also affect WTP for a PSG study. Thus, future studies are needed to investigate the effect of these variables on WTP for PSG studies.

We used multivariate analysis to evaluate the effect of independent variables on WTP. We did not find that objective PSG variables such as RDI and AI significantly affected WTP. In addition, the fact that the child had undergone adenotonsillectomy did not influence WTP. These details are provided to the physicians; however, these data are transparent to parents. These parameters, in addition to the clinical history and physical examination, enable the physician to explain and suggest to the parents how to proceed with OSAS management. Socioeconomic level did not affect WTP for a PSG study. The relationship between WTP and socioeconomic level among different studies is inconsistent. Ryan et al¹⁷ found that WTP for antenatal care correlated with socioeconomic level. Sorum¹⁸ did not find any correlation between parents' WTP to avoid the events and outcomes associated with acute otitis media and socioeconomic level. It appears that regardless of the socioeconomic level, the child's health status is a high priority of the family. Our findings need further reinforcement, since we indirectly measured socioeconomic level, ie, according to clusters of settlements and not by household income. The bid, age times the bid, and the subjective variable health status significantly influenced WTP.

How valid is the contingent valuation approach for estimating WTP for health benefits?³³ The WTP exercises involve hypothetical expenditures rather than the respondent's actual purchasing decisions. Several studies^{36,37} have tested the criterion validity of the WTP method by comparing the stated WTP to the respondents' actual WTP. These studies have shown that the stated WTP exceeds actual WTP. Loomis et al³⁶ showed that stated and actual WTP can be made more credible by improving the survey design.

We arbitrarily selected the age difference of 6 years because it represents a reasonable age span. In our previous study,¹⁰ we found that children under 5 years of age have more-severe OSAS than do children over 5 years of age, and their healthcare utilization is maximal. Most children in our study were younger than 8 years old. Since younger children are heavy consumers of healthcare resources, we believe that their parents are more concerned and, therefore, are expected to have a higher WTP for a PSG study. Further support for these conclusions is the outcome that parents of children who underwent adenotonsillectomy were approximately 1.8 times more likely to pay any given sum than were parents of children who had not undergone adenotonsillectomy. Why do the subjective and objective variables such as health status influence

WTP for PSG study? Subjective parameters such as history and objective parameters such as otolaryngologic examination and audiotape snoring have been shown to have low sensitivity and specificity for the diagnosis of OSAS.^{1,7} Screening studies have limited diagnostic value and, thus, may be useful for initial testing if PSG studies are not available. Polysomnographic study is necessary to assess OSAS severity and to determine treatment.¹ The sleep specialists, pulmonologists, and otolaryngologists in our settings routinely provide this message clearly, which was probably reflected by the parents' expectations of the benefits resulting from the PSG study.

Cost-benefit analysis uses WTP estimation with the contingent valuation approach for evaluating public benefit from the health service. Our results show a benefit of \$572 per diagnosis. According to the concept developed by Drummond et al,¹¹ this benefit should be considered an approximation rather than an exact value, and eliciting preferences should be seen as more useful for group than for individual decision making.^{18,38}

SUMMARY

Performing PSG studies for children with OSAS is beneficial. According to public opinion, decision makers and sleep specialists are on solid ground using WTP to prioritize allocation of scarce resources for PSG studies.

REFERENCES

- Marcus CL. Sleep-disordered breathing in children. *Am J Respir Crit Care Med* 2001;164:16-30.
- Ali NJ, Pitson D, Stardling JR. Sleep disordered breathing: effect of adenotonsillectomy on behaviour and psychological functioning. *Eur J Pediatr* 1996;155:56-62.
- Bar A, Tarasiuk A, Segev Y, Phillip M, Tal A. The effect of adenotonsillectomy on serum insulin-like growth factor-I and growth in children with obstructive sleep apnea syndrome. *J Pediatr* 1999;135:76-80.
- Gozal D. Sleep-disordered breathing and school performance in children. *Pediatrics* 1998;102:616-20.
- Gozal D, Pope Jr DW. Snoring during early childhood and academic performance at ages thirteen to fourteen years. *Pediatrics* 2001;107:1394-9.
- Tal A, Leiberman A, Margulis G, Sofer S. Ventricular dysfunction in children with obstructive sleep apnea: radionuclide assessment. *Pediatr Pulmonol* 1988;4:139-43.
- Suen JS, Arnold JE, Brooks LJ. Adenotonsillectomy for treatment of obstructive sleep apnea in children. *Arch Otolaryngol Head Neck Surg* 1995;121:525-30.
- De Serres LM, Derkay C, Sie K, et al. Impact of adenotonsillectomy on quality of life in children with obstructive sleep disorders. *Arch Otolaryngol Head Neck Surg* 2002;128:489-96.
- Tarasiuk A, Simon T, Tal A, Reuveni H. Adenotonsillectomy in children with obstructive sleep apnea syndrome reduces health care utilization. *Pediatrics* (in press).
- Reuveni H, Simon T, Tal A, Elhayany A, Tarasiuk A. Health care services utilization in children with obstructive sleep apnea syndrome. *Pediatrics* 2002;110:68-72.
- Drummond MF, O'Brien B, Stoddart GL, Torrance GW. *Methods for the Economic Evaluation of Health Care Programmes*. 2nd ed. Oxford: Oxford University Press; 1997.
- Rothfuss J, Mau W, Zeidler H, Brenner MH. Socioeconomic evaluation of rheumatoid arthritis and osteoarthritis: a literature review. *Semin Arthritis Rheum* 1997;25:771-9.
- Brooks RG. Cost-benefit analysis of patients treated at a rheumatism centre. *Ann Rheum Dis* 1969;28:655-61.
- Robinson R. Cost-benefit analysis. *BMJ* 1993;307:924-6.
- Pätälä H, Niemelä P, Laurinkari J. Cost-benefit analysis of synovectomy of the knee. *Scand J Rheumatol* 1976;5:227-32.
- Johannesson M. *Theory and Methods of Economic Evaluation of Health Care*. Dordrecht, The Netherlands: Kluwer Academic Publishers; 1996.
- Ryan M, Ratcliffe J and Tucker J. Using willingness to pay to value alternative models of antenatal care. *Soc Sci Med* 1997;144:371-80.
- Sorum PC. Measuring patient preferences by willingness to avoid—the case in otitis media. *Med Decis Making* 1999;19:27-37.
- Stavem K. Willingness to pay: a feasible method for assessing treatment benefits in epilepsy? *Seizure* 1999;8:14-9.
- Gordon KE, Dooley JM, Camfield PR, Camfield CS, MacSween J. Treatment of febrile seizures: the influence of treatment efficacy and side-effect profile on value to parents. *Pediatrics* 2001;108:1080-8.
- Reuveni H, Schweitzer E, Tarasiuk A. A cost-effectiveness analysis of alternative at-home or in-laboratory technologies for the diagnosis of obstructive sleep apnea syndrome. *Med Decis Making* 2001;21:451-8.
- Israel Law Book No. 1469. National Health Insurance Act; 1996:156-207.
- Brouillette RT, Hanson D, David R, et al. A diagnostic approach to suspected obstructive sleep apnea in children. *J Pediatr* 1984;105:10-4.
- Slothuus U, Brooks RG. Willingness to pay in arthritis: a Danish contribution. *Rheumatology* 2000;39:791-799.
- Bishop RC, Heberlein TA. Measuring values of extra market goods: are indirect mea-

- asures biased? *Am J Agricultural Econ* 1979;61:926-30.
- Boyle K, Bishop R. Economic Benefits Associated with Boating and Canoeing on the Lower Wisconsin River. *Economic Issues* No. 84, Department of Agricultural Economics. Madison: University of Wisconsin; 1984.
- Bowker J, Stoll J. Use of dichotomous choice method to value the whooping crane resource. *Am J Agricultural Econ* 1988;70:373-81.
- Hanemann M, Loomis J, Kanninen B. Statistical efficiency of double-bounded dichotomous choice contingent valuation. *Am J Agricultural Econ* 1991;73:1255-63.
- Statistical Abstract of Israel. No 53. (Tables 12 and 37). Central Bureau of Statistics. State of Israel; 2002.
- Grados Y, Blushtein-Livnon R. Sociopolitical Atlas of Beer-Sheva. The Negev Center for Regional Development. Beer-Sheva, Israel: Ben-Gurion University of the Negev; 2001.
- Currie GR, Donaldson C, O'Brien BJ, Stoddart GL, Torrance GW, Drummond MF. Willingness to pay for what? A note on alternative definitions of health care program benefits for contingent valuation studies. *Med Decis Making* 2002;22:493-7.
- National Oceanic and Atmospheric Administration. Report of the NOAA panel on contingent valuation. *Federal Register* 1993;58:4601-14.
- Bala MV, Mauskopf JA, Wood LL. Willingness to pay as a measure of health benefits. *Pharmacoeconomics* 1999;15:9-18.
- O'Brien B, Gafni A. When do the "dollars" make sense? Toward a conceptual framework for contingent valuation studies in health care. *Med Decis Making* 1996;16:288-99.
- Gylmark M, Morrison GC. Demand for health care in Denmark: results of a national sample survey using contingent valuation. *Soc Sci Med* 2001;53:1023-36.
- Loomis J, Brown T, Lucero B, Peterson G. Improving validity experiments of contingent valuation methods: results of efforts to reduce the disparity of hypothetical and actual willingness to pay. *Land Econ* 1996;72:450-61.
- Neill HR, Cummings RG, Ganderton PT, Harrison, GW, McGuckin T. Hypothetical surveys and real economic commitments. *Land Econ* 1994;70:145-54.

APPENDIX 1

A) Survey to families whose child is scheduled for PSG study – group 1.

Hello, is this the _____ family? May I speak with the father/mother (circle who you talked to)? "My name is _____ and I'm calling from the Sleep-Wake Disorder Center in Soroka University Medical Center. May I please have your attention for few minutes? I'm running a survey of children suspected of having obstructed sleep apnea syndrome (OSAS). Your child is suspected of having OSAS and is currently scheduled for nocturnal sleep study. As you already know, some children with clinical signs suggesting OSAS require sleep studies to establish the diagnosis. If an OSAS diagnosis is confirmed, several therapeutic approaches will be offered to minimize the associated effects. Our survey examines the benefits of expanding sleep laboratories in the southern region of Israel. As you know, the cost of diagnosis is covered by the Clalit Health Care Services insurance as part of the basket of services determined by law. This is an anonymous survey and none of your personal details will be disclosed in my research. I need your help in order to evaluate your utility from the diagnosis. Try to consider a few parameters in your answer: The expected improvement in your child's health, the expected improvement in your family's quality of life, your budget limitations, and the opportunity cost of time spent performing the study. To reiterate, I emphasize that the diagnosis is covered by your insurance and your response may serve to improve this service.

- In order to evaluate your utility from the diagnosis, please let me know if you are willing to pay _____NIS for PSG diagnosis? (Yes/No)
- Could tell me to what extent you are worried about your child's health?
 - I'm not worried at all.
 - I'm a little worried.
 - I'm worried enough to turn to a doctor.
 - I'm very worried.
 - I think that my child's life is in danger.
- How many children sleep with your child in his room? _____
- How many children do you have? _____

B) Survey to families whose child underwent a PSG study but didn't undergo T&A – group 2.

Hello, is this the _____ family? May I speak with the father/mother (circle who you talked to)? My name is _____ and I'm calling from the Sleep-Wake Disorder Center in Soroka University

Medical Center. May I have your attention for few minutes? I'm running a survey of children who suffer from obstructive sleep apnea syndrome (OSAS). The survey examines the benefits of adding OSAS diagnostic services in sleep laboratories in southern Israel. As you probably know, this diagnosis is covered by the Clalit Health Care Services insurance. I need your help to evaluate your utility from the diagnosis that your child underwent. This is an anonymous survey and none of your personal details will be disclosed in my research. We know that your child recently underwent a PSG study and was diagnosed as having OSAS. In light of this knowledge and the recommended therapy, please consider a few parameters in your answer: the improvement in your child's health, the improvement in your family's quality of life, your budget limitations, and the opportunity cost of time spent performing the study. To reiterate, I emphasize that the diagnosis is covered by your insurance and your response will may serve to improve this service.

1. In order to evaluate your utility from the diagnosis, please let me know if you were willing to pay _____ NIS for this diagnosis. (Yes/No)
2. Could you tell me to what extent you have felt relief since your child underwent the PSG diagnosis?
 - I have felt no relief
 - I have felt a little relief (not significant)
 - I have felt some relief
 - I have felt significant relief
 - I have felt significant relief and I'm not worried for my child's life
3. How many children sleep with your child in his room? _____
4. How many children do you have? _____

C) Survey to families whose child underwent PSG and T&A – group 3

Hello, is this the _____ family? May I speak with the father/mother (circle who you talked to)? "My name is _____ and I'm calling from the Sleep-Wake Disorder Center in Soroka University Medical Center. May I please have your attention for a few minutes? I'm running a survey of children who suffer from obstructive sleep apnea syndrome (OSAS). The survey examines the benefits of adding OSAS diagnostic services in the sleep laboratory in southern Israel. As you probably know, this diagnosis is covered by the Clalit Health Care Services insurance. I need your help to evaluate your utility from the diagnosis that your child underwent. This is an anonymous survey and none of your personal details will be disclosed in my research. We know that your child recently underwent both a PSG study and surgery. In light of your personal experience, please consider a few parameters in your answer: The improvement in your child's health, the improvement in your family's quality of life, budget limitations, and the opportunity cost of time spent performing the study. To reiterate, I emphasize that the diagnosis is covered by your insurance and your response may serve to improve this service.

1. In order to evaluate your utility from the diagnosis, please let me know if you are willing to pay _____ NIS for this study? (Yes/No)
2. Could you tell me to what extent have your child's health and your family's quality of life improved since undergoing the diagnosis and the surgery?
 - There was no improvement
 - There was a little improvement (not significant)
 - There was some improvement
 - There was a significant improvement
 - There was such a significant improvement in my child's health that I think that my child's life was saved
3. How many children sleep with your child in his room? _____
4. How many children do you have? _____