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CLINICAL REVIEW

Health care costs and the sleep apnea syndrome☆

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Summary This paper reviews some concepts on health economics from the authors personal perspective. It then examines the few papers published on health economics analysis applied to the field of sleep apnea syndrome, as well as the literature on the indirect (cost) consequences of sleep apnea syndrome. It appears that undiagnosed sleep apnea leads to a roughly two-fold increase in medical expenses in the years preceding the diagnosis and that treating the disease (once it is diagnosed) results in a decrease in these excess costs. It seems clear that sleep apnea increases the actual number of road traffic accidents, which will carry a definite, but unmeasured up to now, economic cost consequence. From the health economic point of view, the best diagnostic strategy is the one with the greater utility (i.e. polysomnography), although it could appear at first sight to be the more expensive one. From the patient's perspective, sleep apnea results in a given decrease in the possibilities to enjoy life, and its treatment is worth considering, especially if one takes into account that the actual treatment costs are not great. The global image of the health costs related to sleep apnea is still blurred, and further work is required to get the complete and clear picture of the economic consequences of this disease and of its treatment.

Introduction

The meaning of the word 'health' depends on the general context of the discourse where it is used. It is certainly not the same for a physician, for an economist or for a person with a cold, an open fracture of the tibia or with a severe chronic invalidating disease. It has become fashionable to consider health, in economic terms, as a commodity among others, with neither less nor more hierarchy or importance than energy (electricity, gas and so on), sports or sugar. It is therefore reasonable, *from that point of view*, to analyze the economic flows related to a given commodity, like health, using the ultimate comparison criteria, which is money.

If we accept these premises, then we can consider health as comprising a series of activities, equipment, salaries, consumables, that represent a given use of economic resources leading to certain results, and analyze whether the economic resources have been used in a valid or efficient way to attain the results, also called outcomes.

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Thus, we can compute overall costs and benefits, calculate cost-effectiveness ratios, cost-benefit ratios, cost-utility ratios, and so on.

It is essential for a physician to recall that nothing of the above has anything to do with truth, neither with biology or medicine. The economic discourse we are referring to reflects the dominant philosophy of our society for the time being, which is essentially materialistic with money as the main endpoint and reference.

Economists sustain that economic resources are inherently scarce, and that the attribution of resources to one end implies by necessity that those resources can't be allocated to other ends, so that one needs some external reference scale (i.e. money) to compare the results or outcomes of the attribution of resources to several different realms in order to make meaningful comparisons and reach 'best' decisions. We will take some lines to consider these statements. To say that economic resources are *scarce* is similar to saying that the amount of matter available for transformation into energy inside the sun is *not infinite*. It is a fact so obvious that one may omit to realize that the fact in itself has no consequence whatsoever. It is a truth in absolute terms, but it conveys no practical consequence of any kind. However, if the essential difference between 'not infinite' and 'scarce' or 'limited' is not made, then we are faced with a problem. If resources are indeed limited, then they cannot be wasted, and have to be managed with care and prudence. Then, and only then, the allocation dilemma becomes true, and we need to compare in some way the efficiency of the use of the scarce resources as they are attributed to one or the other end (ends which by definition are competing between each other for the scarce resources) in order to make the best informed choices and avoid wasting those valuable scarce resources. Enters thus the manager. Indeed, if choices have to be made, someone has to make them, ideally someone skilled at the complexities of measuring and comparing the efficiency (or the inefficiency) of the use of resources. If the best way to compare the use of resources is to establish an external absolute reference scale; if that scale is the economic value attributed to the particular resource we are considering and to the particular outcomes we choose, then the obvious manager is the economist. Is the sun making the best possible use of its scarce and valuable mass to obtain the energy we need for life on Earth? And if the answer is 'no', can we allow the sun to go on wasting that vital matter? Of course not. Some intervention will have to be decided to stop the dangerous and intolerable attitude of the sun. In other words, saying that resources are scarce does *not necessarily* imply that they are limited *to the point where an intervention is desirable or necessary*. If we forget these subtle but important points, then the fact that resources are not infinite leads inescapably to the need of managers, scales, measurements and economists. To make this point as clear as possible: resources are scarce by definition, so that they are scarce everywhere, in USA as in Belgium. The USA spends 14% of its gross national product (GNP) in health to get a certain result. Belgium spends 8% of its own GNP in health to obtain another result, which some consider better.[1] Is the USA spending too much, or Belgium not enough? If the USA cuts down its spending in health to 8%, will that figure be reasonable, or will it still be excessive? And if the USA cuts its spending to 8%, should then Belgium cut its own spending to 5%? And what about the UK, or Italy (7.6 and 8.4%, respectively)? If resources are really scarce, what is the aim we should tend to? 3%? 0%? And who should decide? The absurdity of this debate is obvious.

Of course, we could choose a completely different scale and outcomes. Scales and outcomes are a matter of choice, and are not determined beforehand by Mother Nature. We could well choose suffering as the external scale, and happiness as the outcome. How much human suffering is needed to obtain a certain amount of happiness? Let us suppose that one could estimate, using appropriate measuring instruments, the amount of human suffering needed to produce either a given amount of coal, or an equivalent amount of gas. And that one could also estimate, using other appropriate instruments, the amount of happiness per capita linked to heating during winter months in cold climates. One could perfectly well decide that the amount of human suffering needed to obtain a given amount of gas that will result, by its burning and consequent heat production during winter, in a given amount of happiness for a certain number of human beings in cold climates, is higher than the amount of human suffering required (to attain the same amount of happiness for the same number of human beings) to obtain the same level of heat production from the burning of coal. And that therefore, coal has a higher happiness/suffering ratio than gas, and that therefore coal is to be preferred to gas *even if it costs more money*.

Now we can come back to health economics, but with the above caveats present in our minds, to review what is known on the economic aspects of the sleep apnea syndrome. This means, in simple and schematic terms, to consider the costs of the undiagnosed disease both for patients suffering from the disease, their families and friends, and for society as a whole. The costs of diagnosing the disease and treating it should then be taken into account. For this, one should compare the costs of different diagnostic strategies (for instance using cost/utility ratios), and the costs of different treatment modalities (for instance using cost/effectiveness ratios). One should also take a look at whether diagnosing and treating the disease has any measurable consequence on the costs due to the disease (i.e. economic benefit). A comparison of the economic aspects of sleep apnea and of other diseases should be made. Only thereafter the conclusion will emerge: is sleep apnea a disease with enough economic impact to merit the interest of society and the allocation of resources for its treatment? Or should we forget about it and turn to more appealing issues like diabetes, halitosis, cancer or athlete's food? We will try to examine these issues in the rest of this paper. The available literature is rather scarce so that the conclusions that could be reached will necessarily consist of approximations rather than certitudes.

The costs of sleep apnea

The costs of a disease can be classified in direct and indirect costs. Direct costs are for instance those related to payment of the physician, of a hospital admission, or of drugs required for treatment. Indirect costs can include absences from work, reductions in earning capacity, costs of illness-related accidents and so on (non-medical indirect costs). They can also include direct medical costs related to complications of the undiagnosed disease. These can be called indirect medical direct costs. Once the situation is serious enough that the patient seeks medical help for the specific disease we are interested in, direct costs will begin to accumulate during the process leading to a diagnosis. This process can be more or less simple and lengthy, depending on the characteristics of the disease and the awareness of it within the medical profession at a given place and time. The cost will also depend on the direct costs of the technical procedures needed to establish a firm diagnosis, whether they require an admission to hospital for specific tests, whether they require very specialized personnel et cetera. In addition, these diagnostic procedures may also imply indirect costs (days off work for diagnostic procedures). If there are iatrogenic complications of the diagnostic tests, these might also imply direct and indirect costs.

Indirect medical direct costs related to sleep apnea

In the case of the sleep apnea syndrome, most of the above information is simply not available. Three studies have analyzed the global health care costs *in the years before diagnosis* in a cohort of patients suffering from sleep apnea and compared them to those of a control group reasonably matched for confounding factors. As far as this implies the assumption that the eventual cost difference between patients and controls depends solely on the presence of a disease process called sleep apnea syndrome, but are not strictly related to its diagnosis or treatment, these health-related costs can be included in the indirect medical direct costs of the disease.

Kapur et al.[2] explored the health care costs of 238 adult patients with sleep apnea living in Washington State, USA, during the year preceding the formal diagnosis of the disease. They compared these costs to those of a group of 476 age and sex matched subjects without sleep apnea but enrolled in the same health care program as the patients. The results showed that patients incurred significantly higher mean and median health care costs than controls during the year preceding the diagnosis of sleep apnea. Costs for patients were about two-fold the costs for controls. This remained true after adjustment for the 'chronic disease score', a global measure of chronic disease status. Interestingly, patients had a more severe score than controls (essentially because of hypertension and coronary artery disease, depression, bronchial asthma and diabetes) but, as stated, this did not explain the cost differences. Moreover, there was a significant direct relationship between sleep apnea severity (assessed through the apnea-hypopnea index, AHI) and health care costs, so that the latter increase rapidly as the AHI increases up to a value of 30. Further increases in the AHI resulted in much lesser increases in health care costs. The authors calculated that the health-related cost burden for undiagnosed sleep apnea in the USA is US$ 3.4 billion per year. The main problem with this study is that there was no matching between patients and controls in terms of body weight. Since obesity is common in patients with sleep apnea, the health care cost difference between patients and controls could be due to obesity, and not to sleep apnea, if controls were to be leaner, which is unknown.

Kryger and coworkers have explored in two papers the health care utilization in Canadian patients with sleep apnea during the 10 years preceding, and the 2 years following, the diagnosis of sleep apnea. In 181 patients and 690 age, gender and postal code matched controls, mean individual costs for patients were significantly higher than those for controls (about a two-fold difference) during the 10 years preceding the diagnosis, and these cost differences increased along the years approaching the time of diagnosis, especially for the last 3 years prior to diagnosis.[3] The excess costs include both physician claims and hospitalizations. This study, as the preceding one, has not performed matching for body weight, so that the differences might be due not to sleep apnea, but for instance to obesity. It is not clear from the presented data whether the costs of the diagnosis are included in the calculations, which would of course artificially increase the indirect medical direct costs for patients in the last year prior to diagnosis, by adding to them the direct medical costs related to the disease.

In their second study, Kryger and coworkers followed the health-related costs in 344 male Canadian patients diagnosed with, and treated for, sleep apnea.[4] A total of 1324 controls without a diagnosis of sleep apnea were matched for age, gender and postal code to the patients (3–4 controls per patient). They studied the costs during the 5 years preceding, and during the 2 years following, the diagnosis of sleep apnea. The costs related to the diagnosis itself (sleep laboratory evaluation) were attributed to the first year *after* the diagnosis, to avoid artificially increasing the costs of the last year before diagnosis. Costs related to CPAP equipment were not considered. For follow-up purposes, patients were divided among those complying with treatment (mainly CPAP therapy, $n = 282$) and those not complying ($n = 62$). The data show that patients incur higher absolute costs than controls before diagnosis (somewhat less than two-fold), and that these absolute costs decrease after diagnosis, but there is no statistical evaluation of these figures. The authors analyzed the significance of the cost difference between patients and controls. This shows a significant reduction in the difference between patients and controls from the last year before diagnosis to the second year after diagnosis and treatment of sleep apnea. Indeed, the differences were significant only for patients complying with treatment, but not for non-compliant patients. However, costs remained higher for patients than for controls (no statistical analysis provided). Again, as in the previous study, differences include both physician visits and hospital stays. In the last year before diagnosis, physician-related costs were $260 higher per patient, and this was reduced to $174 per patient in the second year after diagnosis for the entire patient group. The analysis according to compliance with treatment shows the following figures: compliant patients spent in physician-related claims $267 more per patient than their own controls during the year before diagnosis, and $181 during the second year of treatment. Non-compliant patients spent $236 more per patient than their own controls during the year before diagnosis, and $141 during the second year of follow-up (all costs are in Canadian $). It is worth mentioning that non-compliant patients were significantly older and had less severe sleep apnea, and showed a trend towards less obesity. Moreover, during the 5 years preceding diagnosis, non-compliant patients had significantly higher costs for circulatory and genitourinary disorders. Hospital stays significantly decreased after diagnosis for the entire group of patients. The decrease was significant for patients compliant with treatment, but not for patients not complying with treatment. This study is difficult to interpret, because many variables are not available. It is impossible to relate this study and the previous one, since absolute values were not compared in this study, and differences were not compared in the previous one. Obesity was taken into account in neither study. We are told that non-compliant patients do not decrease the difference in costs with respect to their controls after diagnosis, but this difference is less (though we are not told if significantly so) in absolute terms in non-compliant than in compliant patients. We know that non-compliant patients had more circulatory and genitourinary problems than compliant patients before diagnosis, but we do not know whether this was also the case for their respective control groups. Finally, the cost of treatment of sleep apnea in patients was not included in the calculations. If this cost is added to the expenses of the patients, the results could well become non-significant, or even offer a reverse picture.

In spite of the weakness of these three studies, it seems that undiagnosed sleep apnea carries an increase in health care costs; that the excess cost is directly related to disease severity at the time of diagnosis; that the excess cost increases with time, and that the excess cost might decrease with an adequate treatment if well adhered to.

More recently, the same group of investigators[5] have assessed in another group of 773 patients with obstructive sleep apnea, and in a control group matched for age, gender, residence and family physician, the reasons for health care costs in the 5 years previous to diagnosis. Health care costs were 23–50% higher in patients, which is less a difference than in the previous studies from the same authors.

However, in this study, the cost of medications were not included in the analysis. The excess costs were due to a higher number of physicians' visits, and to higher physician fees and hospitalizations. Patients were significantly more likely to have comorbid conditions (hypertension, chronic heart failure and chronic obstructive airways disease). However, it is impossible for this study to assess the role of obesity, since on the one hand the BMI of the control group is unknown, and, on the other hand, the authors found no correlation between BMI and health care costs when they grouped their patients in five groups of increasing BMI.

We have voluntarily excluded from this analysis the possible consequences of sleep apnea on hypertension and stroke. We did so because there are no hard and validated data on the effects of treating sleep apnea on the evolution, treatment and costs of hypertension and stroke (although epidemiological data seem to confirm the etiologic role of sleep apnea as one of the factors causing hypertension or stroke).

Indirect non-medical costs related to sleep apnea

Indirect non-medical costs have not been assessed in patients with sleep apnea. Some considerations can nevertheless be made from the studies of traffic and domestic accidents in patients with this disease. Indeed, one of the main symptoms of the sleep apnea syndrome is an excessive level of daytime somnolence. It is plausible that excessive somnolence, by a decrease in attention, could result in a reduction in the ability to react promptly to unexpected events. If excessive somnolence leads to overt sleep under inappropriate conditions (while driving a motor vehicle, for instance), the ability to execute motor tasks disappears, and a motor vehicle crash can ensue. Accidents due to sleep while driving are generally described as involving a single vehicle, with no avoidance reaction, and leading to serious health consequences (very serious injuries or death).

Several studies, some of them of very high scientific quality, have assessed the risk of traffic accidents in patients with sleep apnea, by comparison to a control group or to a country general population risk. Some studies have also assessed the evolution of the traffic accident risk after treatment for sleep apnea is instituted. Similarly, some authors have studied the number of all accidents (both traffic and home accidents) before and after therapy, as well as the number of days in hospital due to accidents and their evolution with treatment.[6-12] Table 1 summarizes the most recent studies and their main results. It is clear from these data that the untreated sleep apnea syndrome results in a certain amount of indirect costs. These include the costs related to the excess accidents themselves (vehicle repairs or replacement, hospital stays, days lost for work), but do not include the unknown costs related to severe injuries or lives lost. Indeed, most studies have assessed accidents not leading to serious injuries or death. It also appears that an adequate treatment reduces these indirect costs.

Some idea on indirect costs related to days off work can be derived from the studies of health care costs. Indeed, hospital stays show a two-fold increase in hospital days before diagnosis in patients compared to controls. Since one can assume that most of these days are week-days, and not week-end days, there is a certain amount of excess workdays lost due to undiagnosed sleep apnea, that should be computed as representing indirect costs for the disease.

However, the amount of indirect costs remains unknown. Estimations of the overall costs of drowsiness related motor vehicle accidents in the USA for 1988 are in the range of US$ 50 billion. However, there is no data on the part of this cost that could be attributed to sleep apnea.

In 2001, George[12] published a study on accidents reduction following the start of a continuous positive airway pressure treatment. Patients ($n = 210$) had a higher number of motor vehicle collisions than a group of control subjects (matched for age, sex and type of driver's license) during the 3 years before starting treatment. The number of collisions normalized during the 3 years after nasal CPAP, whereas it remained high in 27 patients not using nasal CPAP. There was no change in the number of collisions in the control group. It should be stressed that in this group of patients, driving exposure (km per year) was roughly the double of the usual exposure in Ontario. Based on George's data, Findley and Suratt[13] have calculated that treating 500 patients for 3 years would save US$ 1,000,000, 369,000 in direct property damage and medical expenses, and 648,000 in legal and administrative costs. As we will see later, treating 500 patients for 3 years would roughly cost less than US$ 600,000.

Sleep apnea and its treatment: the patient's point of view

Physicians are used to assess the general health status of patients with simple questions like "How are you today?"; "How are you feeling?". The evolution of symptoms under various treatments can be followed

Table 1 Main studies on accidents in sleep apnea.

First author (Ref)	Type of study (n)	Main outcome	Main results				Main adjustement variables
Barbe[7]	Case-control. 60–60	OR for traffic accidents for the 3 yr preceding inclusion	2.3 (0.97–5.38) for single accidents 5.2 (1.07–25.29) for >1 accident				Age, gender, km/yr, alcohol
Krieger[10]	Questionnaire Cohort study, 1 yr before-1 yr after CPAP. 547	All accidents (*n*/100 persons)		*GFP*	*Pr*	*Po*	
			Car accidents	0.4	8.4	2.5	
			Domestic	3.7	4.6	2.4	
			Work	2.1	2.2	1.3	
			Hospital days related to Accidents	na	885	84	
Young[8]	Cohort study, general population. 913	OR for multiple accidents during 5 yr for subjects with AHI > 15 vs. subjects with no sleep disordered breathing	7.3 (1.8 → 25)				Age, gender, miles/yr
Teran-Santos[6]	Case control. 102–152	OR for one traffic accident	6.3 (2.4–16.2) for AHI > 10				Age, gender, alcohol, driving experience, BMI, visual refraction defects, sleep schedule, history of traffic accidents, medications
Cassel[9]	Questionnaire cohort study preCPAP- 1 yr post CPAP. 59	Traffic accidents rate per 100.00 km	0.8 before CPAP to 0.15 on CPAP				
Masa[11]	Interview (4.002) followed by case control (107–109)	OR for crashes in habitually sleepy drivers vs. controls	OR of 13.3 for auto crashes, OR of 8.5 for total respiratory event index > 15 in sleepy vs. non-sleepy drivers				Age, sex, BMI, alcohol and medications, hours driven per month, years of driving
George[12]	Case-control. 210–210	MVC rates for the 3 years before and after CPAP therapy for patients vs. controls	Before treatment: 0,18 (0,29) vs. 0,06 (0,17), after treatment: 0,06 (0,17) vs. 0,07 (0,18)				Age, BMI, AHI, class of driver's license

OR, odds ratio; GFP, general French population; Pr, Pre-CPAP; Po, Post-CPAP; na, non-available; AHI, apnea-hypopnea index. CPAP, continuous positive airway pressure; MVC, motor vehicle crash.

with questions like "How is your daytime sleepiness?"; "Do you still need to nap after dinner?"; "Do you still fall asleep while watching TV or when your mother-in-law comes to your place?". Nowadays, the assessment of the state of the patient by the physician has come under serious suspicion of inexactitude and partiality. Patients may not wish to disappoint their doctor, and the latter may dislike to hear (and write on records) that his treatment is of no value. One way to circumvent this problem is to assess the impact of a disease state (or of a change in the disease state due to a treatment) for the patient through the measurement of utilities and the related concept of Quality Adjusted Life Years (QALY). Let's assume that an individual becomes ill. This may imply a series of nuisances: symptoms that may be more or less painful, the inability to continue performing a number of tasks that the person was used to perform, like drive a car, play football, or go shopping (if we think about a disease that impairs all these abilities). The individual, now called a patient, may follow a treatment that will allow him to recover his previous healthy state. Some patients will want very hardly to receive the treatment, and recover their previous healthy state. Others, perhaps more fatalistic in nature, will accept the treatment with less enthusiasm. Some may even consider that they can adapt happily to the new disease state, and refuse to go back to their previous healthy state. The strength of an individual patient's preference for a given outcome (recovery of the previous healthy state for instance) is called a utility. It depends on many subjective psychological factors, on the age and personal history of the subject, but also on the nature of the disease and its associated symptoms and consequences, and on the nature, secondary effects and risks of the treatment. In one usual way to calculate utilities according to the mathematical 'standard gamble' method, the subject is placed in a choice situation between a given state (for instance his current health status for the rest of his expected life), and a gamble between two other states, generally 'full health for a given number of years' and 'immediate death'. The probabilities of the gamble are varied by steps until the patient becomes indifferent to the choice between the gamble and the alternative (which is generally, as we have stated, his current health state). At that point it may be considered that, if full health represents an utility of 1 and immediate death of 0, the average of the probabilities in the gamble weighted by their respective values of 1 and 0 is equivalent to the expected utility of the gamble and, since the patient is indifferent to the choice, is equivalent also to the utility of the alternative, which is the current health status.[14] Thus, a utility takes into account the preference for a given level of health for a number of years. Other methods used to compute utilities are the time trade-off method, and a series of general or disease-specific questionnaires. Examples of these are the EuroQol or the Functional Outcome of Sleep Questionnaire.

Let us imagine a one-eyed blind individual. If he is a retired sedentary man in good health with a nice visual acuity in his remaining eye, he may well consider that, apart from a slight level of anxiety due to the fact that he has only one eye left, his health problem is not that a big trouble (for instance 0.1 on a scale from 0 to 1), and does not impair his capacity to enjoy life. If asked how many years of his remaining life (let us say 10 years) he is ready to give-up to recover his lost eye, he might answer 'none', or perhaps '1 year'. Consider now a second one-eyed blind individual. He is younger, and is a professional hunter who lost his useful eye for hunting. For this individual, being one-eyed blind might represent a huge health problem (for instance 0.5 on a scale from 0 to 1). If asked how many years of his remaining life (let us say 40 years) he is ready to give-up to recover his lost eye, he might well answer '20 years'. Thus, for the first retired individual, his health problem is valued as 0.1. Since his life expectancy is 10 years, the QALY value is 1 (0.1×10). For the younger hunter, his health problem is valued at 0.5. Since his life expectancy is 40 years, the QALY is 20. This allows to estimate the time-adjusted utility of a health problem (or of a disease) independently from an eventual treatment.

Let us now consider the case of two blind people. A medical intervention gives them back the use of one eye. One of them may consider this a true miracle, valuing it enormously (for instance 0.8 on a scale from 0 to 1). The other may go on the rest of his life complaining because only one eye, and not both, was recovered, and consider the medical intervention as of little value (for instance 0.2 on the same scale). If both individuals have a life expectancy of 10 years, the medical intervention will be worth 8 QALY's for the first individual, but only 2 QALY's for the second one. This allows therefore to estimate the time-adjusted utility of a given treatment.

Tousignant and coworkers[14] studied the QALY value of CPAP therapy in 19 patients with sleep apnea, by comparing the utilities given by the patients to their health status under therapy and before diagnosis. They found that CPAP added an average of 5.39 QALYs. The range was large, from 0 to 28, reflecting the wide variation in the subjective value given by the patients to the change in their health status due to the treatment of their disease. One problem with this study is that it was a retrospective study: the utilities were in fact measured after CPAP treatment had been instituted for a number of years. Therefore, the utilities reflected a comparison between 'actual conditions' (relatively easy to assess) and 'previous conditions' (requiring both a recall process and an imagination effort). Chakravorty et al.[15] have confirmed that nasal CPAP resulted in a 23% improvement in the baseline severely impaired health status, adding 8.2 QALYs to a group of 37 patients treated with CPAP, whereas the improvement in a control group treated with 'lifestyle counseling' but not with nasal CPAP was of 4% with an addition of 4.7 QUALYs (significantly less than in the nasal CPAP group). These data were obtained using the standard gamble approach. Interestingly, when the EuroQol method of assessment was used, no significant differences were noticed between the nasal CPAP and the 'lifestyle' groups. The authors did not conclude that there were no differences, but that the EuroQol questionnaire was not appropriate to assess a sleep apnea population. This exemplifies one problem with questionnaires: if they do not detect the expected differences, it is rapidly concluded that they should be replaced with other, more sensitive, instruments, able to show what one wanted to show.

Mar et al.[16] have recently performed a similar study. Utilities were assessed prospectively in 46 newly diagnosed patients, both before and after 3 months CPAP therapy, using the EuroQol 5D instrument. The mean age of the patients was 53 years, whereas the AHI was 41.3/h. The BMI was $39.7 + 13.6\ kg\ m^{-2}$. This is probably a very obese sample, more than what is generally seen in recently published studies. The mean gain in EuroQol score was 0.073 units (somewhat less than found by Tousignant et al.), whereas the baseline score was 0.738.

Tousignant et al.[14] have also calculated the cost-utility ratio of CPAP therapy. To do this, they tried to take into account all costs related to life-long treatment, including CPAP as well as a single therapeutic polysomnography per patient. They related then this total cost to the total number of QALYs gained with the treatment, and obtained thus the cost per year of QALY added. The figure varied between Canadian $ 9.800 and 3.500 per QALY, depending on the assumptions made to calculate costs. To get an idea of what this means, the reader might like to know that a coronary artery bypass surgery for a left main coronary artery occlusion has a cost-utility ratio of US$ 6.200 per QALY. Renal dialysis costs US$ 47.000 per QALY, and screening asymptomatic patients for carotid stenosis US$ 120.000 per QALY.[17] Mar et al.[16] in their recent paper, using their own calculated utilities, confirm the cost-utility ratio of €7.800 per QALY if calculations are made with a time span limited to 5 years, and €4.938 per QALY if calculations are made on the basis of the lifespan. They also performed sensitivity analysis, and only in the worst possible case did the cost-utility ratio exceed €20,000 per QALY.

Direct medical costs: the diagnostic tests for sleep apnea

This is perhaps the right point to introduce the reader not familiar with health economics in the complexities of this realm. Classically, a patient with a clinical suspicion of sleep apnea should undergo a diagnostic procedure to confirm the suspicion, and only then receive treatment. A diagnostic procedure could consist in the reference test (the gold standard, full night polysomnography), or a 'validated' (not reference) test, like ambulatory respiratory polygraphy. Some people contend that giving a CPAP trial is all that is needed to identify patients with sleep apnea responsive to CPAP, and that no diagnostic test is necessary in these patients. Only those patients with a clinical suspicion of SAS and not happy with the CPAP trial would need a diagnostic work-up. If one thinks on these three strategies in economic terms, it rapidly becomes apparent that the reference test strategy will be the most expensive. The CPAP trial would probably be the cheapest (after all CPAP machines are not disposable, and if a patient brings back one machine you could still use it for the next patient), whereas the ambulatory polygraphy strategy would appear as intermediate in cost. However, if one introduces the utility concept into this seemingly easy economic situation, things may look otherwise.

A single study has evaluated the cost-utility characteristics of three diagnostic strategies for assessing patients with a clinical suspicion of sleep apnea. The three chosen strategies were full night polysomnography followed by CPAP therapy in patients with a confirmatory test; an unattended home cardiorespiratory sleep study followed by CPAP therapy in patients with a positive result, and a treatment CPAP trial with no diagnostic test in all patients clinically suspected of sleep apnea.[17] According to the sensitivity and specificity characteristics of the three strategies, a number of patients without sleep apnea (false positive patients) will receive CPAP therapy, and a number of patients with sleep apnea (false negative patients) will be denied treatment. All data used in this model study were obtained from the literature. The utility data were those of Tousignant et al.[14] Sensitivity and specificity characteristics of the three strategies were obtained from different published sources. Costs were computed from the charges for the different visits, tests and treatment as practiced in the University of Michigan Sleep Center, which are quite higher than costs in Europe. To avoid criticisms concerning the paucity and the uncertainty of the published data and assumptions, the authors submitted their model to a wide sensitivity analysis. Using a Monte Carlo simulation, all baseline variables were simultaneously allowed to vary between reasonable limits (for instance, the average utility for CPAP in a patient with sleep apnea, which is 0.87 according to reference 14, was allowed to vary between 0.4 and 1), and the resulting cost-utility ratios compared between the three strategies. Cost-utility ratios were expressed in QALYs for the first 5 years after the initial testing (QALY5), to take into account that most published data on compliance, survival, et cetera concern a time span of around 5 years. The results showed somewhat surprisingly that the more expensive strategy (i.e. full night polysomnography) resulted in the best cost-utility ratio: 4.019 QALY5 for full night polysomnography, 3.955 for unattended home study, and 3.934 for clinical-based decisions. The incremental cost-utility ratio for polysomnography compared to home studies was US$ 13.431 per QALY, whereas this value was US$ 9.165 for the comparison between polysomnography and clinical-based decisions. The results were not sensitive to wide variations in the baseline characteristics, except for extreme and highly unlikely ones. The reasons why the more expensive strategy yields the best cost-utility ratios seem to depend not only on the sensitivity and specificity characteristics of the three strategies (favoring full-night polysomnography, which was considered as the gold standard), but also on the frequency of positive findings in the population to be tested, and on the fact that diagnostic costs are 'relatively' low compared with treatment (or no treatment in false negative patients) costs. Diagnostic errors have thus a high cost consequence. Mar et al.[16] have recently confirmed these conclusions. In their own study, using a cardiorespiratory polygraphy study for diagnosis, and a split night polysomnography to confirm diagnosis and titrate nasal CPAP, was compared to the use of full night polysomnography both for the diagnostic and for the separate titration nights. The increase in cost related to the second, more expensive, approach, changed little the incremental cost-effectiveness ratio, that remained below €13,000 per QALY for a prevalence of the disease in the tested sample that was allowed to vary from 0.32 to 0.16.

Direct medical costs: treatment costs in sleep apnea

There are no published studies assessing the costs of sleep apnea therapy. Several reports have made cost considerations of auto-CPAP therapy used as a diagnostic and therapeutic tool as compared to standard polysomnography and CPAP treatment, but they are flawed by simplistic and not validated assumptions. Nowadays a simple constant CPAP device with a built-in clock memory to assess compliance, which is all that is needed in the very vast majority of patients,[18] costs about US$ 800 (or less) both in Europe and the USA. The life expectancy of one of these devices is about 5–10 years. Assuming a discount rate of 4%, and assuming that all treatments are effective (a reasonable assumption for polysomnographic titrated CPAP in sleep apnea) the total cost is US$ 865 for a machine with a life expectancy of 5 years (US$ 173 per year), or US$ 950 for a life expectancy twice as long (US$ 95 per year). Consumables (one standard mask per year, one tubing every 2 years, one standard head-gear per year, filters) amount to approximately US$ 100–200 per year. Thus the total 'pharmacological' cost for the treatment of a patient with sleep apnea with the best available therapy is something around US$ 300 per year, to which the cost of electricity, neglected here, is to be added. For most patients, it is unusual to need more than a single annual compliance visit, and new polysomnographies are certainly not required as standard practice. Thus, the total cost including physician fees is in the range of US$ 350. Mar et al.[16] have recently calculated an annual cost of €358 in Spain, a very similar value. For comparison purposes, the pharmacological cost for the treatment in Belgium of one patient with simple systemic hypertension, with for instance an ACE inhibitor and a diuretic, is about US$ 360 per year. For a patient with angina pectoris, treated with a beta-blocker, aspirine, a nitrate derivate and an ACE inhibitor, the cost is in the range of US$ 570 per year. A patient with mild to severe chronic obstructive pulmonary disease with frequent exacerbations treated with inhaled corticosteroids and long-acting β_2 agonist, inhaled anticholinergics, two courses of antibiotics and oral corticosteroids per year will incur pharmacological costs of US$ 954 per year. An important point to bear in mind is that the cost of treatment for a patient with sleep apnea is independent of the severity of the disease, which is not the case for other diseases.

The cost of other treatment modalities cannot be estimated so simply, both because they vary too much among countries and practitioners, and because their effectiveness (success or failure rate) has to be taken into account to distribute the cost of the procedures failing to relieve sleep apnea among the ones succeeding in relieving the disease. Calculations for UPPP should for instance take due notice that the success rate is well below 50%, and that even when successful, the disease can recur after some years.

Discussion

We have tried to review all available literature on the health economics of the sleep apnea syndrome. The first impression that comes to mind is that this source of information is not vast, is not complete, leaves too many aspects unexplored, and is frequently written in a language that is not accessible to most physicians. Economic analysis does not follow simplistic assumptions, and uses specific mathematical methodologies that are in general well beyond the skills and background of most medical practitioners. Health economics is a dangerous game, that we are forced to play even if we ignore its rules.

Once this has been said, some comments can nevertheless be made. If one considers only the very convincing data on *non-medical* indirect, traffic accidents-related, costs due to sleep apnea, it is certain that this disease is worth considering.

However, the magnitude of this effect is difficult to establish. Imagine for instance the possibility that the nuclear Chernobyl accident was due to a sleepy technician with severe sleep apnea unable to react adequately to a system warning. If this was true, then all costs due to the Chernobyl accident should be included in the indirect costs of the disease, making it one of the most expensive diseases worldwide! If Chernobyl had nothing to do with sleep apnea, its indirect economic weight would be much less impressive, but could still be high enough to consider that diagnosing and treating the disease is economically profitable. Indeed, treatment results in a measurable reduction in traffic accidents. What is not known is whether the cost of diagnosing and treating the disease is directly profitable in economic terms, in the sense that it could lead to a reduction in indirect costs due to accidents in *some* patients with sleep apnea (those who had accidents!) that exceeds the costs of diagnosis and treatment in *all* patients with sleep apnea.

As far as indirect medical direct costs are concerned, the data are less convincing, but suggest that the undiagnosed disease leads to a doubling in medical expenses with respect to a control population. This increase in medical expenditures is related to the severity of the disease, and is independent from the (non-sleep apnea related) chronic disease status of the patients. Treatment of the disease results in a measurable reduction of the difference in medical expenditures between patients and controls. The magnitude of this reduction (and even its mere existence) is, however, not well established. Again, we do not know whether this reduction in indirect medical costs with treatment offers an economic 'benefit' (the investment is recovered in excess), or merely reduces by a certain amount the excess in indirect medical costs.

If we consider the patient's point of view, it appears that the disease results in a certain decrease in the possibility to enjoy life, and that an adequate treatment is worth considering. This is an everyday clinical experience, but has been rigorously assessed only twice, and in mostly severe or very obese patients. One may wonder whether extending the analysis to less severe or obese patients could alter the picture, and if the results would still show a beneficial value for CPAP.

The diagnostic strategy analysis suggests that, at least in this specific disease, not making diagnostic errors is of prime importance in economic efficiency terms, so that the more expensive strategy is the one with the higher cost-utility ratio.

The treatment cost analysis, though based on the author's personal impressions in Belgium and not on serious validated data, makes sleep apnea a rather inexpensive disease to treat.

A last comment is worthwhile concerning costs. Costs are very variable from country to country. A polysomnography in most European countries costs a third or less than in the USA. Thus, when conclusions are derived from the literature, it should be remembered that they cannot be 'translated' in economic terms to other countries or settings. Caution is essential if huge mistakes are to be avoided.

How can we relate all this to the clinical world? Sleep apnea is a fascinating disease, because it can seriously hamper the joy of life for the patient himself, his family, his friends and his employers, and because a treatment exists that allows almost instantaneously, by turning a button, to recover normal sleep and, most importantly, normal wakefulness. From the medical point of view, there is absolutely no doubt that this disease is worth diagnosing and treating. Where, then, lies the problem? The problem is to desperately try to justify this *in economic terms*. Because if one does not, then the managers might decide that the disease, respectable as it may be in medical terms, is not worth considering from the financial point of view. Indeed, there are many medical conditions that may make life miserable, but that do not carry serious economic consequences (like having a disgraceful nose, halitosis and so on). The manager may be sympathetic to the individuals suffering from these conditions, but he will not allow the use of scarce economic resources to take due care of them. He will rather devote those scarce resources to treat coronary heart disease, which has been rigorously proven to cost a lot of money, and whose treatment has been shown to be profitable in economic terms. Or may be not.

And one comes back to the real problem: are resources really scarce? Or are we just told that they are? The answer is neither medical nor economical nor political. The answer is philosophical. For the time being, we will have to play the game of health economics. Let us learn the rules.

Practice points

- Sleep apnea results, roughly, in a two-fold increase in the medical expenses in the years preceding the diagnosis.

- Treating the disease results in a measurable decrease in these excess costs.
- Untreated sleep apnea increases domestic, workplace and road accidents. The costs related to these accidents have not been evaluated.
- The diagnostic strategy with the greater utility is not necessarily the less expensive one.
- From the patient perspective, sleep apnea decreases the ability to enjoy life, and its treatment is worth considering.
- The cost of CPAP treatment appears as reasonable.

Research agenda

- The cost consequences (both medical and non-medical) of accidents related to sleep apnea should be measured.
- The costs of CPAP treatment should be rigorously assessed.
- The cost-effectiveness ratios of the various available treatment modalities (CPAP, surgery, mandibular advancement devices) should be calculated.
- Utility analysis in sleep apnea should be extended to patients with mild and moderate forms of the disease.

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* The most important references are denoted by an asterisk.