

Costs of disease: The perspective matters

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Costs of disease: The perspective matters

For many centuries, care for the ill was provided within families. For the poor and those without family, charitable organizations took over this task. At the turn of the 18th century, healthcare became slowly a public responsibility. In parallel, debates emerged how to finance healthcare. The Bismarckian and Beveridge systems served as the best-known examples which were introduced in the 19th century. Independent of the type of financing, the costs of diagnosing and treating illness became increasingly a key factor in organizing healthcare.¹ Since then, studies on the costs of disease have become increasingly common. "Costs" usually refers to the financial consequences of disease, although in some studies (typically costs-of-illness [COI] studies) the loss of health-related quality of life is also considered a cost ("intangible costs"), yet rarely expressed in monetary terms. Although the usefulness of studies investigating the costs of disease has been debated frequently,²⁻⁷ knowledge of costs can inform the decision-making process on healthcare and social policies, and is useful to prioritize areas for future economic evaluation. Moreover, data on costs serve as a main component of cost-effectiveness studies.^{4,8-10} However, in this setting of healthcare policies, the part of the costs of disease borne by patients and their families are not explicit and receive relatively little attention. Notwithstanding, financial impact of diseases on patients and families can be large and influential on social status and consequently on health behavior and health.

For correct interpretation of cost studies, it is essential to understand their concept and limitations. Cost studies are no economic evaluations: the costs are not weighted against health or other benefits, and there is no estimation of how costs might change with the introduction of a new intervention or change of policy. These studies also do not provide an indication of health: some diseases result in high individual suffering but only a limited utilization of resources. The impact on quality and quantity of life should therefore always be taken into account by policy-makers to put costs in a correct perspective.¹¹ Finally, although cost studies can be used to prioritize disease for economic evaluation or identify the most important cost components of a disease, they do not provide an indication of efficiency or waste of resources. Diseases associated with high costs might be interesting for policy-makers when prioritizing healthcare, but might not be preventable or treatable, or disease management might come at an inevitable high cost.

Cost studies, and health economic studies in general, can be conducted from several perspectives.¹² The adopted perspective

guides which cost categories are included in the analysis, with the societal perspective being the most comprehensive as it includes all cost components, inside and outside of healthcare.¹³ For this reason, health economists often prefer this perspective. However, costs can also be assessed from the specific perspective of the patient (Table 1). Patients and their families often incur significant disease-related costs. These can be related to healthcare use (out-of-pocket direct costs), non-healthcare uses (travels to care providers, over-the-counter drugs, informal care) but also to indirect costs caused by impaired working ability (loss of income and employment, early retirement, reduced career opportunities). The amount of costs borne by patients are relevant, as high costs might restrict patients' access to successful treatment or even lead to health inequalities and poverty in the long-term, which in itself has an effect on poor health.¹⁴

1 | A RECENT EXAMPLE - COSTS OF GOUT FROM THE PATIENT PERSPECTIVE

In this issue of *International Journal of Rheumatic Diseases*, Nathan et al. aimed to estimate the direct and indirect out-of-pocket costs of gout to Australian patients.¹⁵ In their population study, 79 patients with gout completed an open, web-based questionnaire, which contained questions on their resource use and work productivity in the last 12 months. The median (mean; range) total direct out-of-pocket costs were estimated at AU\$200 (AU\$666; AU\$0-AU\$7088) per year. Medical costs, especially those related to medication and consultations with care providers, accounted for the majority (>99%) of these direct costs. The median (mean; range) number of missed work days was 0.25 (3.2; 0-65) in the last 12 months, which was valued at AU\$60. Some patients were unable to afford medical care for gout, in particular medication. Nathan et al. need to be commended for their efforts of investigating a relevant aspect of chronic illness. Formally not a COI study (as intangible costs were not included), their study estimated both direct and indirect costs from the patient perspective. As such, it instigates a discussion on the costs of disease from the perspective of patients and their families, and specifically on the role of the indirect costs associated with short- and long-term absence from work (sick leave, work disability, early retirement) and (more arguable) of presenteeism (ie, decreased productivity while working).

**TABLE 1** Overview of the costs borne by society and patients for different cost categories

Category ^a	Societal perspective	Patient perspective
Healthcare, such as therapies, diagnostics, drug prescriptions	"True" cost (independent of who pays for them, this includes patients' co-payments)	Only the part of the costs paid by patient or their family (contributions, co-payments)
Non-healthcare, such as travel costs, membership to society, over-the-counter drugs, informal care	Market price	Market price
Other sectors, such as costs related to paid work	Cost resulting from reduction in production of goods and services	Income loss as a consequence of ill health (eg, in case of sick leave or work disability, or if career prospects are affected)

^aHealthcare and non-healthcare costs are often called "direct costs" as they are directly related to health services. Healthcare refers to costs borne by the healthcare system, while non-healthcare refers to costs borne by parties outside the healthcare system. Costs in other sectors are not directly related to health services, and are called "indirect costs".

2 | ESTIMATING PRODUCTIVITY COSTS - CHALLENGING, BUT NECESSARY

Productivity changes related to paid work can be due to absenteeism (being absent from work) or presenteeism (decreased at-work productivity). For absenteeism, a distinction is made between temporary absenteeism, also referred to as sick leave, and permanent absenteeism (work disability, early retirement). Productivity changes are an important aspect of the costs of disease. This is especially true for chronic diseases that manifest themselves in patients of working age, for which the consequences of such diseases on work outcomes can span multiple decades. The relevance of productivity changes for society has been highlighted in cost studies of rheumatic diseases (including gout), where these costs of productivity changes accounted for substantial parts (often > 50%) of total costs.¹⁶⁻¹⁸ The far majority of cost studies in gout adopted either a societal or payer perspective, while studies on the burden of productivity costs for patients with gout are mostly lacking.¹⁶

2.1 | Assessment of productivity changes

Measuring changes in "production" as a consequence of illness is a challenging but essential first step to estimate indirect costs for patients and society, and even recommended in guidelines on health economic studies.¹⁹ To assess loss of "production of good and services", the *time (days, hours)* a person is not able to work is usually regarded as an appropriate surrogate of true loss of production. Next, a *person's wage* is commonly used as an appropriate monetary value of lost production and converts time into costs. In the study by Nathan et al., the authors have chosen to use survey data and respondents had to indicate "the number of (half days) days absent because of gout in the past 12 months".¹⁵ As they correctly state, longer assessment periods to detect relatively rare events (sick leave) will improve precision when the sample size is small. Also, while longer periods of recall are prone to inaccuracy, in clinical studies a balance has to be made between frequency of measurement, and study design, feasibility and patient burden.^{20,21} Absence of specification in the survey whether calendar or work days should

be counted, might add to inaccuracy. For short spells, persons might count work days missed, while in case of long-term absence patients might find it easier to estimate the calendar days absent. Also, patients might find it difficult to decide whether sick leave is attributable to gout or other health problems, and this is especially relevant when the prevalence of comorbidities is high (as in gout and thus in this study). An employed gout patient admitted 2 weeks to hospital because of worsening heart failure, who experienced during his stay a gout attack lasting 4 days, how would this be reported in a survey asking to report days absent due to gout? Or what about the 2 days sick leave because of colchicine induced diarrhea? To avoid such discussions and to allow benchmarking with the general population, it is advised to collect data on absence related to any health problem. To complicate measurement of loss of production related to sickness absence, when a gout flare is short and a patient returns to work after 2 or 3 working days, it is likely colleagues took over the most urgent tasks during normal working hours and the patient catches up on unfinished tasks himself upon return. These so-called compensation mechanisms might reduce production loss of short-term sickness toward zero. Of note, they could also result in more stress (eg, due to increased workload at times), which can be considered intangible costs and are often not quantified in monetary terms.

Assessment of *presenteeism* comes with additional challenges, which are as of yet not fully resolved, and is a topic of continuing research. First of all, it is important to recognize differences in the underlying concepts among instruments assessing presenteeism. Some of these focus on the consequences for society (productivity/efficiency), while others mainly aim to measure the patient's experience (difficulty/inability) when performing work and have no (primary) economic objective.^{22,23} Patients in this survey were asked to rate the extent to which gout problems affected their productivity, as a surrogate for the percentage of "productive" time loss.^{24,25} Alternatively, researchers could have asked to rate the extra hours patients would have needed to catch up unfinished tasks.²⁶ To estimate the indirect costs, the % productive time lost (or the extra time to catch lost production) can be converted into monetary terms, again using wages. Clearly, these different approaches result in very different estimates of productive time lost and thus indirect costs.^{22,27} Although a gold standard is lacking, the extra time



needed to catch up on lost work is a more conservative approach, avoiding overestimation and reducing the risk of stigmatization of persons with a chronic disease.²⁸ Having said this, the jury of health economists is not out whether presenteeism results in any loss of production (and indirect costs) at all, or whether the greater majority of a person's presenteeism is compensated. Whatsoever, in the current study, Nathan et al. recognized they did not ask patients to indicate the number of hours they would have normally worked (in a week), preventing them from calculating the working time lost due to presenteeism. Even if that information would have been available, recall of presenteeism should be accounted for when appraising the results. Presenteeism likely fluctuates strongly over time, and it is unlikely patients can estimate their averaged presenteeism over the past year.

2.2 | Productivity changes – who pays the price?

Apart from general measurement issues in relation to accuracy, the most important issue when estimating indirect costs from a patient perspective, is the question which costs matter in this perspective. While restrictions to participate in the work process will likely impact health-related quality of life, there is not necessarily a cost impact for the patients or their families. This would only be the case if (a) wage is affected in case of sick leave or work disability, (b) when recurrent sick leave would affect career prospects and thus future income, or (c) when free (unpaid) time is used to catch up the unfinished work. It would be interesting to know whether in this study the reported days of sick leave indeed resulted in reduced income, and whether the study might underestimate income loss of employment or early retirement because of health. To answer these questions, insights into social security and income compensation in Australia, and how income is affected in case of sick leave opposed to work disability, would be helpful. Knowledge on such elements of a study's context benefits the interpretation of productivity costs borne by patients, and it should be encouraged to report them. If income loss, in particular due to unemployment and work disability for medical reasons, was indeed underestimated in this study, it is easier to understand the economic hardships reported by a substantial proportion of patients (33%). This underlines the importance of careful assessment of *all* aspects of productivity: the productivity costs borne by patients and their family are real and do affect their health and welfare.

3 | CONCLUDING REMARKS

Chronic disease often has economic consequences for patients and their families. These consequences go beyond costs for medical care and medication. Patients are at risk of losing income and future earnings, which can put substantial strain on households. These costs can also affect access to care, putting patients and their families in a vicious circle. Appropriate assessment of productivity

changes, valued in the context of the relevant social security regulations and income compensation measures, is necessary to correctly appreciate the extent of costs of disease borne by patients. Research on costs of gout and their consequences from the patient perspective, of which the study by Nathan et al. is a first step,¹⁵ is necessary to identify where and how to support these patients and break the circle.

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