Review Article

Cost Of Illness Studies For Disease Burden: A Review

Usha Adiga[1*]

[1*]Professor, Nitte-DU, KS Hegde Medical Academy, Department of Biochemistry, Mangalore, India, E-mail: ushachidu@yahoo.com

***Corresponding Author**:- Dr Usha Adiga

*Professor, Nitte-DU, KS Hegde Medical Academy, Department of Biochemistry, Mangalore, India, E-mail: ushachidu@yahoo.com
DOI: 10.47750/pnr.2022.13.S08.491

Abstract

In most economic and business decision-making processes, whether private or public, the term "cost" is always taken into consideration, whereas its opposite can range from benefit (in cost-benefit analysis, CBA), to effectiveness (in cost-effectiveness analysis, CEA), and particularly in healthcare fields, to quality-adjusted life years (QALYs), or latent utility (in cost-utility analysis, CUA). Even though they are occasionally evaluated in different ways, costs are normally measured in monetary terms to enable a simple comparison of competing options. A phrase used to represent the various ways in which a disease impacts the health of a country, a region, a community, or even an individual is cost of illness (COI), also known as burden of disease (BOD).

The term "COI" refers to a number of factors, including the frequency or prevalence of a disease, its effects on longevity, morbidity, and the subsequent decline in health status and quality of life (QoL), as well as financial aspects, like direct and indirect costs related to early death, disability, or injury caused by a specific disease and/or its comorbidities. Accurate knowledge of COI is crucial since it aids in the development and prioritisation of healthcare treatments and policies, as well as the eventual distribution of healthcare resources in accordance with financial restrictions to achieve policy effectiveness. Understanding how the costs are recognised, categorised, and quantified in the COI research is the aim of this effort because it is essential to do so.

BASIC CONCEPTS OF COI STUDIES

The main goal of the COI study is to assess the financial burden that sickness has on society as a whole. According to Jefferson et al., the goal of COI research is descriptive: to compile, appraise, and sum the costs of a particular circumstance in order to give a broad picture of its financial burden (1). Therefore, when conducting COI studies, researchers must acknowledge, characterize, catalogue, measure, and give a value to the costs that an illness and its comorbidities may entail. According to Clabaugh and Ward (2008), "Analyzing COI affords useful chances for communicating with the public and policy makers on the relative relevance of certain diseases and injuries" (2).

It's crucial to keep in mind when conducting COI research that the economic COI is supposed to represent the "possible benefits of a health care intervention" if it had effectively treated the disease. Accordingly, the COI studies often include some form of "health loss" indicator and try to put a number on the resource costs involved in treating the related diseases. The BOD studies clearly evaluate the "weight" of years of life lost (YLL) due to premature mortality and years lost due to disability or illness (YLD).

These two categories make up the total DALYs (disability-adjusted life years), which includes health care costs and the "lost economic or societal contribution" brought on by premature death or disability.

Types of costs

COI studies frequently divide costs into three categories: direct, indirect, and intangible costs. Because intangible costs haven't been quantified in COI research very frequently due to measurement concerns and related arguments, we here largely focus on the first two cost categories.

Direct costs

The direct costs, which are borne by the healthcare system, society, families, and individual patients, encompass both healthcare-related and non-healthcare expenses. The former is referred to as the medical care expenses for diagnosis, treatment, and rehabilitation, among other things, whereas the latter is connected to the usage of non-healthcare resources including transportation, household costs, moving, property losses, and informal cares of any sort. Estimates of the direct expenses of chronic diseases are higher than those of acute illnesses or communicable diseases.

Indirect costs

In COI studies, the term "indirect" occasionally refers to productivity losses caused by morbidity and mortality, which are borne by the individual, family, society, or the employer. This is in contrast to accounting and most business disciplines where the term "indirect" costs indicate the supporting and overhead activities that need to be shared among the users. It has been advised to replace the word with "productivity losses or productivity costs" to eliminate any confusion or uncertainty readers may experience. In actuality, indirect costs account for a portion of the social welfare

losses brought on by illnesses, with the remainder losses being accounted for by the reductions in healthy time brought on by pain, suffering, and bereavement brought on by illnesses.

Different approaches of COI studies

Prevalence- vs. incidence-based approaches

The approaches employed in COI research can be classed as prevalence- or incidence-based depending on how the epidemiological data are used. The first method—the one that is most frequently used—estimates the financial burden of a condition over a set time period, usually a year, while the second method—which takes into account any new cases that arise during that time period—estimates the lifetime costs of a condition from the time it first manifests until it goes away (typically by cure or death). The annual incidence of fatalities and hospitalizations attributable to diseases, as well as the costs associated with such incidences, are calculated through prevalence-based studies (plus other costs such as prevention, research and law enforcement costs).

Incidence-based studies determine the overall number of new hospitalizations or fatalities in a specific year and give each of these new occurrences an estimated lifetime cost. Due to the nature of long-lasting diseases, which necessitates lengthy follow-up periods, the prevalence-based approach is the only viable method for quantifying long-term conditions (1). However, it might not be able to determine how long the conditions will last (3). As a result, prevalence-based techniques often examine the COI in the present and past in a particular year, whereas incidence-based research typically estimates the present and future COI in a given year. For chronic health and social issues like the use of illegal drugs, the results of prevalence-based and incidence-based estimates are usually equivalent.

For health conditions that are decreasing in size, prevalence-based estimates will likely be less precise than incidence-based estimates. Incidence-based estimates often provide bigger estimates than prevalence-based estimates for emerging health crises like hepatitis infection epidemics because many infected people may still be in the latency phase of the diseases. Tarricone outlines the situations in which each tactic is more useful and appropriate (4,5). The prevalence-based method can be particularly beneficial when the study's main objectives are (4,5)

1. To draw attention to conditions whose impact has unintentionally been underestimated by those in charge of developing health policy. Due of the numerical differences between the two methods, the prevalence strategy is able to achieve this objective more successfully than the incidence-based method.
2. To create policies that reduce costs. This is so that decision-makers have a broad understanding of the burden as a whole and, more crucially, the main cost components, or the areas where cost containment measures would have the biggest impact.

The incidence-based strategy is particularly useful when the following study's primary objectives are:

1. When taking preventative measures into account, incidence-based techniques assess the potential savings that could result from the preventative measure's adoption.
2. Examining the whole illness management strategy. With this strategy, researchers can create clinical and therapeutic recommendations meant to improve the overall and each individual stage of the illness management process.

Prospective vs. retrospective approaches

Depending on the relationship between the start of the study and the data collection, COI investigations can be carried out either prospectively or retrospectively. In a retrospective approach, all important events have already occurred at the time the study is initiated, and we are only collecting already recorded data. Contrarily, with a prospective strategy, the pertinent events have not yet happened at the start of the study, therefore data collection must be carried out by monitoring the patients over time. Studies based on incidence and prevalence of COI can be conducted either prospectively or retrospectively (5). Because all key events have already happened, the main advantage of a retrospective method is that it is less expensive and time-consuming than its alternative so that they can be measured and recorded in a dataset.

The retrospective method can only be effective in examining a disease with a long history if there are enough observational datasets available. However, in a prospective strategy, analysts should be able to create the data collection methods they wish to use. The analysts can create a comprehensive dataset on the sickness and the use of medical resources at every action and intervention from the questionnaires created and given to patients, family members, and/or medical professionals. A prospective and incidence-based method would be very costly and time-consuming when investigating a disease over a lengthy period of time; retrospective COI studies may be more effective at determining the burden of sickness.

TOP-DOWN VS. BOTTOM-UP VS. ECONOMETRIC APPROACHES

1.Top-down approach

The top-down approach, known as the epidemiological or attributable risk approach, measures the proportion of a disease that is due to exposure to the disease or the risk factors. Developed by Morganstern et al. (1980), this approach uses aggregated data along with a population-attributable fraction (PAF) known as epidemiological measure to calculate the attributable costs (6-9).

2) Bottom-up approach

The cost estimation can be divided into two parts when using a bottom-up approach. Estimating the unit costs of the inputs used to create and provide particular medical and health care services is the second step after measuring and

quantifying the health inputs that were used in the first. Unit costs are multiplied by the quantities utilized to determine the overall expenses. The main challenge here is that depending on the study's scope, different data will be required and available. National level survey datasets are frequently utilized because they offer trustworthy information on medical care utilization, allowing researchers to fairly quickly estimate the quantity of medical care services as well as unit costs or pricing values. We can avoid the possibility that the entire cost of treating individual diseases exceeds national health expenditure (NHE) in a given country by using the comprehensive research to distribute total national spending among the major diagnostic categories. But applying the NHE may cause the total direct costs to be either underestimated or overestimated, so the top-down method is likely to result in incorrect cost allocation. Additionally, since various disease categories may absorb different non-health expenditures, the removal of cost categories from NHE accounting such as transportation and informal care distorts estimates of costs by disease category. Additionally, this approach has a fundamental flaw in that all costs are solely assigned to the initial diagnosis if we consider that a relevant part of all hospital discharges involve patients with multiple diagnoses (5).

3) Econometric approach

The econometric strategy is to calculate the cost disparity between a cohort with the disease and a cohort without it. The two cohorts are matched by numerous demographic and mediating characteristics, other chronic illnesses, and typically through a series of regression studies. There are two main techniques for estimating costs that fall under the econometric approach: a mean differences approach and a multiple-stage regression approach. The incremental difference attributed to the disease is calculated using a comparison of the mean expenses experienced by each of the two cohorts using the mean differences approach (10,11). Studies that employ the mean differences approach may only give the disease's per-case cost rather than its total cost.

If there are many cases with zero costs and a small number of cases with extremely high costs, a multiple-stage regression method is often used. The regression estimates with the disease dummy variable deleted and the regression coefficients from the regression analyses with the disease dummy variable included are compared to determine the incremental cost of the disease. Although there are numerous other possible modifications for each regression analysis based on the types of datasets and the research objectives, the regression analysis frequently employs a two-stage procedure to estimate COI (12-15). Since the econometric approach only needs one dataset because it calculates the incremental difference between those with the disease and those without it, which is understood as a strength in adopting this approach(10-14). While the top-down approach usually requires cost data as well as on the relative risks which are needed to calculate the PAFs, the bottom-up approach often requires data from multiple sources for the unit cost and utilization rate of the different types of health care. Though each method is valid and appropriate in some situations, in most cases the econometric approach has more advantage in requiring less data.

Perspectives of COI studies

The COI studies may be conducted from many angles, and each one may contain somewhat different cost factors, which could ultimately result in a wide range of outcomes for the same sickness. These viewpoints could assess the financial burdens on a community, the healthcare system, third-party payers, industry sectors, the government, as well as participants and their families (16-18). However, each viewpoint offers helpful data concerning the expenses incurred by the specific group. The wider social perspective is generally favored because a condition's effects do not just affect the people or organizations who are directly affected. We can identify "cost shifting" between sectors and take into consideration alternative resources used outside the health care sector by using the societal perspective (19-20). The societal perspective is recommended for potential cost analyses like CBA, CEA, and CUA because it allows a thorough analysis of all the opportunity costs attributable to a disease and is the most comprehensive because it includes all direct medical costs and indirect costs for all members of a given society where they are involved (21).

To put it another way, the majority of health economists' efforts over the past 30 years have been focused on either pointing out how disconnected COI studies are from the widely accepted field of welfare economics or identifying connections between COI and WTP methods that could lend credibility to COIs (22). According to this theory, COI studies might become more relevant if they are viewed from a different angle, even though they are of utmost importance in helping to comprehend the major drawbacks of the two approaches. Because it essentially does not compare costs with outcomes, COI analysis differs from other other economic evaluation analyses. It is a descriptive study whose main objectives are:

1. to determine the financial cost of disease to society. This would provide data on the amount of resources that are consumed due to illness, and along with epidemiological information on morbidity and mortality, may help 1. rank diseases according to their global burden, 2. identify the key cost components and their incidence over total costs, and 3. rank diseases according to their local burden.

2. To ascertain the clinical care of illness at the national level. Managers and policymakers would find it simpler to assess how inputs and/or intermediate services are integrated to form the ultimate output, which might range from a single product, such as a hospital stay, to a thorough treatment pattern that incorporates a variety of medical services. The basis for re-engineering the entire process in the event of inefficiencies, as well as for re-evaluating the clinical strategy in the case of evidence-based medicine, can then be laid by bringing forward inefficient and/or ineffective functions. Clinical recommendations may be one of the consequences in this situation when the clinical management of the sickness is determined to be ineffective.

3. to elucidate cost variability In this case, statistical analysis can be utilised to ascertain whether there is any relationship between cost variability and elements related to the illness (such as severity), the patient (such as demographic

information), or the healthcare providers (e.g. teaching hospitals versus district hospitals). These findings would assist managers in providing the planning process with more precise information regarding the future provision of services, as knowing the cost drivers that at least partially explain the consumption patterns of services can be very helpful when planning the provision of health care services. To accomplish these objectives, COIs must be bottom-up and the top-down approach must be unequivocally rejected.

CONCLUSION

COI categorically positions cost analysis as a useful instrument for healthcare sector decision-making. In fact, estimates of the COI based on various methods have many applications. The assumption that policies targeting a disease and its associated diseases should be given top priority in the establishment of policy agendas is typically supported by cost estimates, to start. Each COI study should be assessed against a standard of excellence to which the public is entitled. Without such a requirement, supporters are more prone to emphasize and frequently overstate the COI results by incorporating extra components. Cost estimates also help to focus attention on specific problems and rules. Knowing whether a particular disease of concern has higher societal and/or medical costs is essential. The improvement of cost estimates serves as a reliable baseline measure of the efficacy of health programmes, initiatives, or any other types of interventions that aim to reduce or eliminate the negative effects of disease. Estimates of societal costs can also help in comparing the many approaches that can be used to alleviate the effects of sickness across different countries.

REFERENCES

1. Jefferson T, Demicheli V, Mugford M. Cost-of-illness studies, elementary economic evaluation in health care. 2nd Ed., London: BMJ Publishing Group, 2000:17-29.
2. Clabaugh G, Ward MM. Cost-of-illness studies in the United States: a systematic review of methodologies used for direct cost. Value Health 2008;11:13-21.
3. Kortt MA, Langley PC, Cox ER. A review of cost-of-illness studies on obesity. Clin Ther 1998;20:772-779.
4. Tarricone R. Valutazioni economiche e management in Sanita. Milan: McGraw-Hill, 2004. 23.
5. Tarricone R. Cost-of-illness: what room in health economics? Health Policy 2006;77:51-63.
6. Morgenstern H, Kleinbaum DG, Kupper LL. Department of Epidemiology and Public Health, School of Medicine, Yale University, New Haven, CT. Int J Epidemiol 1980;9:97-104.
7. Hogan P, Dall T, Nikolov P; American Diabetes Association. Economic costs of diabetes in the US in 2002. Diabetes Care 2003;26:917-932.
8. Hodgson TA, Cohen AJ. Medical care expenditures for diabetes, its chronic complications, and its comorbidities. Prev Med 1999;29:173186.
9. Reynaud M, Gaudin-Colombel AF, Le Pen C. Two methods of estimating health costs linked to alcoholism in France (with a note on social costs). Alcohol Alcohol 2001;36:89-95.
10. Birnbaum HG, Kessler RC, Lowe SW, Secnik K, Greenberg PE, Leong SA et al. Costs of attention deficit-hyperactivity disorder (ADHD) in the US: excess costs of persons with ADHD and their family members in 2000. Curr Med Res Opin 2005;21:195-206.
11. Swensen AR, Birnbaum HG, Secnik K, Marynchenko M, Greenberg P, Claxton A. Attention- deficit/hyperactivity disorder: increased costs for patients and their families. J Am Acad Child Adolesc Psychiatry 2003;42:1415-1423.
12. Finkelstein EA, Fiebelkorn IC, Wang G. National medical spending attributable to overweight and obesity: how much, and who's paying? Health Aff (Millwood) 2003;Suppl Web Exclusives:W3-219- 226.
13. Yelin E, Trupin L, Cisternas M, Eisner M, Katz P, Blanc P. A national study of medical care expenditures for respiratory conditions. Eur Respir J 2002;19:414-421.
14. Yelin E, Herrndorf A, Trupin L, Sonneborn D. A national study of medical care expenditures for musculoskeletal conditions. Arthritis Rheum 2001;44:1160-1169.
15. Greenberg PE, Sisitsky T, Kessler RC, Finkelstein SN, Berndt ER, Davidson JRT et al. The economic burden of anxiety disorders in the 1990s. J Clin Psychiatry 1999;60:427-43.
16. Rice DP. Cost of illness studies: what is good about them? Inj Prev 2000;6:177-179.
17. Luce BR, Manning WG, Siegel JE, Lipscomb J. Estimating costs in cost-effectiveness analysis. pp. 176-213. In: Gold MR, Siegel JE, Russell LB, Weinstein MC. eds., Cost-effectiveness in health and medicine. New York: Oxford University Press, 1996 ; 173-213.
18. Hodgson TA. Costs of illness in cost-effectiveness analysis: a review of the methodology. Pharmacoeconomics 1994;6:536-552.
19. Byford S, Raftery J. Perspectives in economic evaluation. BMJ 1998; 316:1529–1530.
20. Goetzel RZ, Long SR, Ozminkowski RJ, Hawkins K, Wang S, Lynch W. Health, absence, disability, and presenteeism cost estimates of certain physical and mental health conditions affecting US employers. J Occup Environ Med 2004;46:398-412.
21. Gold MR, Siegel JE, Russell LB, Weinstein MC. Cost-effectiveness in health and medicine. New York: Oxford University Press, 1996.
22. Glied S. Estimating the indirect cost-of-illness: an assessment of the forgone earnings approach. American Journal of Public Health 1996;86(12):1723–8.
23. Koopmanschap MA. Cost-of-illness studies. Useful for health policy? Pharmacoeconomics 1998;14(2):143–8.