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Review Article

Comprehensive Cost of Illness: A Novel Method to Evaluate Economic Burden of Disease in a Super-aged Society

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ABSTRACT: Appropriate allocation of limited healthcare resources has been among the major challenges of making health policy decisions. Cost of illness (COI) is a simple and convenient approach that measures the burden of disease in terms of monetary cost. This method has been criticized because it only covers costs without considering outcomes, and it is unclear who has to pay the cost. However, COI is relatively simple to calculate compared with other methods for economic evaluation, and it has been used for political decision-making. In an aging society, costs of treating diseases that require long-term care keep on increasing. We propose the comprehensive-COI, which modifies the original method to include both the direct costs of long-term care and the family burden of unpaid costs. Consideration of all the aspects of COIs can facilitate more precise evaluation of the economic burden of diseases.

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KEYWORDS: cost of illness study, economic evaluation of health care, economic burden of disease, health economics, health policy

Introduction

In an aging society, disease structure changes and the proportion of chronic diseases that require long-term care keeps on increasing. Consequently, with the increase in healthcare expenditure, appropriate allocation of limited resources will become more important in health policy making. Long-term care (LTC) costs, including family care cost as well as public LTC insurance cost, will increase. LTC costs have often not been included in the calculations of economic burden of disease, because they are difficult to estimate accurately.

Many countries have witnessed an increase in healthcare expenditure, and economic evaluation of diseases has been regarded as an essential element in health policy decisions in recent years. In many countries, life expectancy is increasing. Advancement in pharmaceutical and therapeutic technology is the reason behind such expectations. However, this advancement has also escalated medical expenses. Specifically, biopharmaceutical costs are rapidly increasing compared with that of conventional medicines, although they may be more effective. The importance of microscopic technology assessment, such as cost-effectiveness analysis, has been recognized. In addition, macroscopic economic evaluation also has become more important. To determine the priority of diseases for allocating limited resources, the burden of diseases must be evaluated. Methodologies to measure the burden of diseases while considering the increasing demand for LTC are needed.

Cost of illness (COI) is one of the methods to evaluate the economics of diseases such as quality-adjusted life

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year (QALY)[1] and disability-adjusted life year (DALY).[2] COI calculates direct and indirect costs of economic burden of diseases through cost accounting. Many countries use the COI method for policy assessment because of its simplicity.[3] However, this method is not yet familiar in Japan, and there are only a few studies on this, including our estimations.[4–9]

In COI, LTC cost is not included, and its applicability for diseases with LTC needs is limited; hence, we propose a modified COI method, called comprehensive COI (C-COI), which covers LTC costs also. In this study, COI and C-COI methods will be explained along with some examples.

Outline of COI Method

The COI method was first used in the analysis of mental diseases by Malzeberg,[10] and it was developed by Rice[11, 12] at the National Center for Health Statistics in the USA. Rice undertook successive case studies and promoted formalizing the method. Basically, COI includes both direct cost (DC) and indirect cost (IC), with IC including morbidity cost (MbC) and mortality cost (MtC). DC is the expense of a specific disease, MbC is the loss of labor value during hospitalization or a hospital visit, and MtC is the loss of human capital caused by the early death. The COI method has been used for macroscopic analysis rather than microscopic analysis. In macroscopic analysis, COI can be calculated as a "top-down" method using aggregated statistics of a nation or a society. In countries whose national data are available, it is possible to calculate COI of major diseases. The effect of policies can be evaluated for disease control in monetary terms, and that evaluation plays a part in prioritizing policies.

COI has been widely used because of such characteristics. The COI method has influenced the decision-making of the government in the USA for more than 30 years. For example, Kirschstein et al. reported the result of COI calculation of major diseases to the US Congress.[13] The studies of COI of tobacco-caused diseases in Medicaid by Miller et al. and Wamer et al. were used for the revision of state laws about the tobacco industry.[14, 15] The COI study of trauma by Max et al. motivated the Centers for Disease Control and Prevention (CDC) to launch a trauma center.[16] The Environmental Protection Agency (EPA) calculated COI of diseases related to environmental pollution.[17] In examples outside of the USA, the COI method was used by the National Institute for Clinical Excellence in the UK, the Pharmaceutical Benefits Schema in Australia, and other cited examples.[18] Tarricone reported that COI studies were among the commonest economic studies in healthcare in Italy and abroad, and they were commonly used by organizations such as the World Bank, the World Health Organization, and the US National Institutes of Health.[11]

The COI method is simple to calculate and is straightforward; however, it has been criticized by many researchers for its simplicity. It has been said that the COI method has no clear theoretical background,[11] and "cost" in the COI method is also criticized because it is not clear who bears the cost.[19] In addition, the COI method has been criticized because it concentrates only on cost and does not compare efficacy and utility of the treatment of diseases.[20] The reason why COI studies are widely used in spite of such criticisms is that COI researchers accepted such criticisms and have developed a more comprehensive COI method, including the presentation of study guideline and a framework for the analysis. For example, Hodgson reported the recommendation of a study guideline and framework for COI analysis in 1982.[21] In 1994, Rice insisted in the *Lancet* that COI researchers were bound to specify their methodology and data for third-party evaluation.[22] In Japan, "Guideline for economic evaluation of healthcare technologies in Japan" was published by a research group supported by Health and Labour Science Research Grants for FY 2012.[23]

Methodology of COI Study (1): Acute Diseases

Original COI includes DC, which is the expense of treatment of a disease, and IC, which is the loss of labor value caused by the treatment or early death. This original COI includes neither decrease of quality of life (QOL) nor cost of LTC. Therefore, the target of original COI is acute diseases, where the decrease of QOL or cost of LTC is not the main burden. For example, cancer is such a disease, and it is suitable for COI calculation. Original COI is defined as follows:

$$\mathrm{COI} = \mathrm{DC} + \mathrm{IC}$$
$$= \text{medical expense of a disease} + \mathrm{MbC} + \mathrm{MtC}.$$

Calculation of DC and IC is based on data of patients with the disease, and the calculation method can be divided into the prevalence approach and the incidence approach from the different viewpoints of patients.[24] If COI is calculated based on all patients in a target area (e.g., a country) at a certain point in time, the prevalence approach that needs an estimation of prevalence rate is used. However, estimation of prevalence rate is quite difficult,

and the estimation of patients' conditions needs some model. Alternatively, the incidence approach uses the number of deaths, both inpatient and outpatient for a period (normally 1 year), which is relatively easier to estimate. Therefore, the incidence approach is convenient for the estimation of macroscopic burden of diseases. In this approach, calculated COI is easy to understand. For example, it could explain "the social burden of lung cancer in 2015."

The COI method defines DC as the medical expenses for treatment of a disease. However, it is difficult to estimate medical costs for treatment of a disease in countries that have no universal national health insurance. Because the expenses for treatment of the disease differ among medical institutions, it is quite difficult to estimate a national medical expense of the disease. For example, efforts to estimate DC in the USA encounter such difficulties. In Japan, most treatments are covered by national health insurance, and the government reports the estimations of national medical expenses by diseases. In such countries, it is relatively easy to estimate DC by a top-down method using national statistics.

IC is divided into MbC and MtC. MbC is the loss of labor value which must be sacrificed when treating the disease, and MtC is the loss of human capital caused by early death, which can be determined by calculating one's lifetime labor value. Therefore, both MbC and MtC can be interpreted as the loss of labor value. In countries that have developed national statistics, it is relatively easy to comprehend the numbers of inpatients, outpatients, and deaths categorized by sex and age categories. Thus, MbC and MtC are grouped by sex and age categories for calculations to show the features of each disease.

MbC is the loss of labor value caused by treatment of a disease, and it can be divided into the loss of labor value caused by hospitalization and the loss of labor value caused by outpatient visits. MbC is calculated using the following equation:

$$\mathrm{MbC} = \Sigma_{i,j}(\mathrm{THD} \times \mathrm{LVd}) + \Sigma_{i,j}(\mathrm{TOVy} \times \mathrm{LVd} \times k).$$

In this equation, i is age class, j is sex, THD is the total person-days of hospitalization, LVd is the one-day labor value per person, TOVy is the total person-days dedicated to outpatient visits, and k is coefficient which means some ratio of one day for outpatient visit time ($0 < k < 1$). LVd can be calculated by multiplying one-day average income by employment rate. However, the value of unpaid work must be considered to estimate the burden of unpaid workers like homemakers. Some countries estimate the annual monetary valuation of unpaid work. Even if there is no data about the annual monetary valuation of unpaid work, it can be acquired by surveying total time for unpaid work. LVd and THD are calculated as follows:

$$\mathrm{LVd} = (\mathrm{Iy} + \mathrm{ULVy})/365$$

$$\mathrm{THD} = \mathrm{HPy} \times \mathrm{ALOS}.$$

In this equation, Iy is the annual income per person, ULVy is the annual monetary valuation of unpaid work per person, HPy is the annual number of hospitalized patients, and ALOS is the average length of hospital stay.

MtC is the loss of human capital, and it can be acquired by calculating the lifetime labor value of a deceased person if they were alive until average life expectancy from 0 years old (LE_0). Because of that factor, human capital value is calculated after categorizing by sex and age. Average labor value generally starts in the mid-teens, peaks in age 50-60 and decreases after age 60. For example, consider an average person who dies at age 20. If they were alive, that person could earn the average income of a 20-year-old in the year of death, average income of a 21-year-old in the first year after death, average income of a 22-year-old in the second year after of death, and they could continue to earn an average income each year until LE_0. That person's human capital can be calculated by summing up these average incomes until LE_0. However, care must be taken to evaluate each year's income at present (i.e., the year of death). Future value must be converted to present value. Expected labor value in years after should be converted to present value of labor by discounting with a discount rate as in the following equation:

$$\mathrm{PVL}_n = \frac{\text{Expected labor value n years after}}{(1+d)^n}.$$

In this equation, PVL_n is the present labor value after n years, and d is discount rate. Human capital can be calculated by summing up PVL_n until LE_0. Thus, human capital value of the person who died at T years old (HC_T) can be calculated as follows:

$$\mathrm{HC}_T = \sum_{i=0}^{n} \mathrm{PVL}_n.$$

However, $n = LE_t$.

Hence MtC is calculated as follows:

$$\mathrm{MtC} = \Sigma_{i,j}(\mathrm{HC} \times \text{number of death}).$$

In this way, COI is defined as the total of DC, MbC, and MtC. Consideration must be given to the difference of each cost's proportion according to each disease.

Methodology of COI Study (2): Diseases that Need LTC

The original COI method is suitable for acute diseases, but it cannot reflect the decrease of QOL and burden of LTC. In the case of diseases which need LTC, the burden of family care can be evaluated using the "loss of labor value" concept as well as direct cost of LTC. In a country that has national LTC insurance such as Japan, LTC direct cost (LTCDC) can be calculated by using national statistics. The C-COI method that includes costs of LTC is defined as follows:

$$\text{C-COI} = \text{MDC} + \text{MbC} + \text{MtC} + \text{LTCDC} + \text{FB}.$$

In this equation, MDC is medical direct cost, LTCDC is LTC direct cost, and FB is family burden of LTC. For C-COI, DC (original COI method) is called MDC to distinguish from LTCDC, but it is defined as actual or direct medical expense of a disease. MbC and MtC are also defined in the same manner as in COI. However, C-COI adds LTCDC and FB. LTCDC includes the burden of LTC services, which are purchased through the market. It is also difficult to estimate LTCDC in countries that do not have national LTC insurance. However, in countries with national LTC insurance, LTCDC can be estimated using the data of LTC payments.

FB is also known as informal care cost,[25] and it means the burden of LTC service which is not purchased through the market. To estimate this part of FB, information from family caregivers is needed. There are few national statistics about such family caregivers, so in most countries, researchers need to estimate FB by a bottom-up method based on their own research. Fortunately, the Japanese government started to track such statistics when national LTC insurance was introduced in 2000. FB is defined as follows:

$$\text{FB} = \Sigma_{i,j} (\text{NFC} \times \text{ATCd} \times 365 \times \text{LVh}).$$

In this equation, i is age class of a family caregiver, j is sex of a family caregiver, NFC is the number of family caregivers, ATCd is the average time for care each day, and LVh is the one-hour labor value per person. Here FB is defined as the loss of a family caregiver's labor value considering his/her sex and age (like MbC). Such estimation is known as the opportunity cost approach (OCA). Additionally, there is another estimation method that calculates FB using the average wage of professional caregivers (AWPC). This method is called a replacement approach (RA). FB by RA is defined as follows:

$$\text{FB} = \Sigma_{i,j} (\text{NFC} \times \text{ATCd} \times 365) \times \text{AWPC}.$$

OCA evaluates the loss of a family caregiver's labor using the average income of their sex and age; as a result, it has a risk of underestimation if the person is elderly. RA replaces the loss of family caregiver's labor with AWPC, so the unit cost of care service is fixed. Therefore, it is thought that RA shows the real burden when compared to OCA.

Example of C-COI Method

Here, we present the results of cost estimations of three major diseases (cancer, heart disease, and cerebrovascular disease (CVD)) in Japan using the C-COI method.[9] Fig. 1 shows C-COI of the three major diseases in Japan using OCA in 2008, 2011, and 2014. C-COI of cancer is the largest impact, and cost amounts to slightly less than 10 trillion yen. CVD is the second largest impact, and cost amounts to slightly more than 6 trillion yen. Heart disease is ranked third, with cost amounts to slightly more than 4 trillion yen. However, the breakdowns of costs are different among the three diseases. The cost of care for cancer, which includes LTCDC and FB, occupies only 3.1% of C-COI in 2014; however, cost of heart disease occupies 12.3%, and cost of CVD occupies 52.0% that year. Cancer is an acute disease, and cost is better estimated with original COI. However, original COI method for CVD may underestimate the economic burden of the disease.

The line graph in Fig. 1 shows the average age of death for each disease, which reflects MtC. It is relatively easy to calculate the average age of death if the C-COI is calculated by the data categorized by sex and age. Cancer has the lowest age of death, and the percentage of MtC is relatively high. The reason is that a higher age of death devalues human capital cost.

The calculation of original COI/C-COI enables the visualization of economic burden associated with various diseases with different characteristics and is expected to contribute to the adoption of future health policies in allocating limited healthcare resources.

Discussion

The calculation of original COI/C-COI enables the visualization of economic burden associated with various diseases with different characteristics and is expected to contribute to the adoption of future health policies in allocating limited healthcare resources. This is why COI studies are widely used until today in spite of many criticisms.

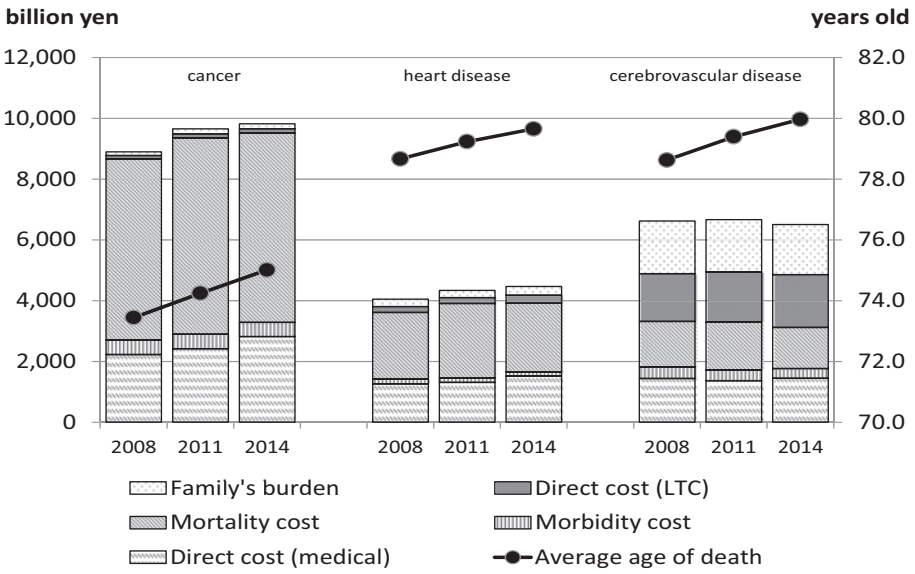

Fig. 1 C-COI and average age of death for three major diseases.
Source: Matsumoto et al.[9]

Moreover, C-COI method enables to evaluate chronical diseases.

However, C-COI method also has its limitations. First, the C-COI method still cannot evaluate QOL accurately. It can only evaluate QOL indirectly using nursing care level. Second, C-COI method cannot be applied to every disease. Some diseases lose patients' productivity even if they work. This is called as "presenteeism." For example, mental diseases have such problems.

COI/C-COI method needs more adjustment for evaluating specific diseases. However, there are new trials for evaluating productivity loss.[26–28] COI/C-COI method still has ample room for improvement.

Conclusion

Despite many criticisms, COI has established its position by securing credibility, repeatability, and comparability rather than modifying its theoretical framework. In an aging society where diseases with LTC will increase, C-COI which includes both direct cost of LTC and FB can evaluate the economic burden of diseases more precisely.

Abbreviations: COI: Cost of illness; LTC: Long-term care; C-COI: Comprehensive cost of illness; DC: Direct cost; IC: Indirect cost; MbC: Morbidity cost; MtC: Mortality cost; THD: Total person-days of hospitalization; LVd: 1-day labor value per person; TOVy: Total person-days of outpatient visits; Iy: Annual income per person; ULVy: Annual monetary valuation of unpaid work per person; HPy: Annual number of hospitalized patients; ALOS: Average length of stay; LE_0: average life expectancy from 0 years old; PVLn: Present labor value n years after; HCT: Human capital value of the person who died at T years old; LTCDC: Long-term care direct cost; MDC: Medical direct cost; FB: Family burden of LTC; NFC: Number of family caregivers; ATCd: average time for care each day; LVh: One-hour labor value per person; OCA: opportunity cost approach; AWPC: Average wage of professional caregivers; RA: Replacement approach.

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