Review Article

A Literature Review of Indirect Costs Associated with Stroke

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Background: Stroke is a leading cause of mortality and long-term disability. However, the indirect costs of stroke, such as productivity loss and costs of informal care, have not been well studied. To better understand this, we conducted a literature review of the indirect costs of stroke. *Methods:* A literature search using PubMed, MEDLINE, and EconLit, with the key words stroke, cerebrovascular disease, subarachnoid hemorrhage, intracerebral hemorrhage, cost-of-illness, productivity loss, indirect cost, economic burden, and informal caregiving was conducted. We identified original research articles published during 1990-2012 in English-language peer-reviewed journals. We summarized indirect costs by study type, cost categories, and study settings. *Results:* We found 31 original research articles that investigated the indirect cost of stroke. Six of these investigated indirect costs only; the other 25 studies were cost-of-illness studies that included indirect costs as a component. Of the 31 articles, 6 examined indirect costs in the United States, with 2 of these focused solely on indirect costs. Because of diverse methods, kinds of data, and definitions of cost used in the studies, the literature indicated a very wide range internationally in the proportion of the total cost of stroke that is represented by indirect costs (from 3% to 71%). *Conclusions:* Most of the literature indicates that indirect costs account for a significant portion of the economic burden of stroke, and there is a pressing need to develop proper approaches to analyze these costs and to make better use of relevant data sources for such studies or establish new ones. **Key Words:** Stroke—economic burden—indirect cost—productivity loss—cost of informal care.
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Stroke, a leading cause of death and long-term disability, is a public health problem worldwide. Globally, there are an estimated 15 million strokes, leading to nearly 5 million deaths and another 5 million cases of permanent disability per year.[1] Because of the increasing size of the elderly population and increasing prevalence of major risk factors, such as hypertension and obesity, stroke is predicted to continuously increase.[2] Moreover, the mortality rates of stroke have kept increasing in some countries in recent decades.[2] Although the United States and some European countries experienced decreasing stroke mortality rates in the same period,[2-4] the decreasing stroke mortality rate and the increasing size of the elderly population increase the long-term disability among survivors of stroke.[5]

Many studies have found high direct costs associated with stroke, including costs for inpatient stays, outpatient visits, rehabilitation, medications, nursing home and so forth. For example, total annual direct costs were estimated at $22.8 billion in 2009 for the United States[6] and €26.6 billion in 2010 for the European Union plus Iceland, Norway, and Switzerland.[7] Far fewer studies have considered the indirect costs of stroke, including productivity loss because of morbidity and mortality and costs of informal

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caregiving usually provided by unpaid family members, although the indirect costs have been claimed to be large.[8]

To better understand the total economic burden of stroke, especially the indirect costs of stroke consisting of productivity loss and informal caregiving costs, we examined peer-reviewed publications of the past 2 decades, including an analysis of the indirect cost. The information we present here will be useful to decision makers in public health and researchers for developing strategies for stroke prevention, treatment, and rehabilitation.

Methods

We performed a comprehensive literature search of peer-reviewed journal articles published in English between January 1990 and September 2012 by using the databases PubMed, MEDLINE, and EconLit. We augmented the search by using Google Scholar and checking the references of the articles we obtained. Key words for the search included *stroke, cerebrovascular disease, subarachnoid hemorrhage, intracerebral hemorrhage, cost-of-illness, productivity loss, indirect cost, economic burden, and informal caregiving*. We investigated 2 main categories of indirect cost: productivity loss and informal care cost. Productivity loss consisted of loss due to premature death (mortality cost) and the cost of disability due to the reduced productivity of survivors of stroke (morbidity cost).[9] The cost of informal caregiving is the value of time spent by family members or other caregivers that is not considered to be part of the care given by formal health care providers.[10] A cost for care provided by formal health care providers such as a home health aide is considered to be a direct cost. Because the proportion of total cost that was represented by indirect cost is a useful indicator measuring the importance of indirect costs estimation, we included cost-of-illness (COI) studies with sufficient analyses of the indirect cost. COI studies estimate the value of all resources spent or forgone, including health care cost and productivity loss, because of stroke.

Figure 1 shows the algorithm used for selecting studies for this review. The initial review of titles and abstracts excluded studies that: (1) were not about stroke; (2) assessed the burden of stroke using nonmonetary terms, such as hours of caregiving or emotional distress; or (3) were only about direct medical costs. In addition, we excluded review articles, editorials, and commentaries. We completed full-text review of all articles that passed the initial review and finalized the set of original research articles for this study by further excluding studies that: (1) were focused on cost-effectiveness; (2) used an unspecified indirect cost for stroke within broad disease categories, such as cardiovascular disease or brain disorders; (3) were about direct costs only, such as studies that included the cost of informal caregiving as a part of direct cost and did not specify indirect costs at all; and (4) were not original studies. We included articles on cardiovascular diseases and brain disorders if the indirect costs of stroke were estimated separately.

We investigated 3 types of study designs. First, we investigated whether a study is a prevalence-based or an incidence-based study. A prevalence-based study examines the costs incurred during a given time period regardless of the date of onset of stroke, whereas an incidence-based study estimates costs of new onset of stroke within a specific period of time for defined lengths of follow-up (lifetime, 1 year, or 6 months).[11]

Second, for estimating the productivity loss, there are 2 approaches: the human capital approach (HCA), which estimates forgone earnings because of stroke as the productivity loss,[12,13] and the friction approach (FA), which assumes a friction cost, a cost associated with the replacement of workers including productivity losses due to substitution of workers or the training costs of new employees, as the productivity loss.

Third, for estimating the cost of informal caregiving, we found 2 methods: the opportunity cost (OC) approach and the replacement approach (RA). The cost of informal caregiving under the OC approach is estimated by using the value of each activity that informal caregivers forgo to provide informal care.[10,14] In contrast, the RA, also known as the proxy good method, assumes that an informal caregiver substitutes for a paid caregiver who would have provided the same type of caregiving services.[10,14]

To compare indirect costs of different countries in different study periods, we derived the 2012 US dollar value by using consumer price indices of study countries in the years of cost analysis and in 2012 from the World Bank and purchasing power parity exchange rate in 2012 from the Organisation for Economic Co-operation and Development.[15]

Results

In all, 31 original articles were selected for our review. Six of them solely investigated the indirect cost,[10,14,16-19] and the remaining 25 were COI studies, which included both the direct and indirect costs (Table 1).[7,9,20-42] Among the 6 studies focusing on indirect costs, 4 investigated the costs of informal caregiving, 1 studied mortality cost, and 1 studied morbidity cost. None of them examined both the productivity loss and the cost of informal caregiving.

As shown in Table 2, which summarized the data sources used to estimate the indirect costs, the US studies relied on national-level survey data, such as the Census data or the National Health and Nutrition Examination Survey, or area-specific surveillance data for the estimation of incidence or prevalence rates of stroke. To estimate the productivity loss or costs of informal caregiving, government data or national-level survey data were used. Non-US studies used various data sources, such as hospital and local area data and national surveys.

Table 3 presents methods and results of the COI studies, 12 prevalence-based studies and 13 studies based