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Burden of epilepsy: A prevalence-based cost of illness study of direct, indirect and intangible costs for epilepsy

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Summary
Objectives: We aimed to gauge the burden of epilepsy in China from a societal perspective by estimating the direct, indirect and intangible costs.
Methods: Patients with epilepsy and controls were enrolled from two tertiary hospitals in China. Patients were asked to complete a Cost-of-Illness (COI), Willingness-to-Pay (WTP) questionnaires, two utility elicitation instruments and Mini Mental State Examination (MMSE). Healthy controls only completed WTP questionnaire, and utility instruments. Univariate analyses were performed to investigate the differences in cost on the basis of different variables, while multivariate analysis was undertaken to explore the predictors of cost/cost component.
Results: In total, 141 epilepsy patients and 323 healthy controls were recruited. The median total cost, direct cost and indirect cost due to epilepsy were US$949.29, 501.34 and 276.72, respectively. Particularly, cost of anti-epileptic drugs (AEDs) (US$394.53) followed by cost of investigations (US$59.34), cost of inpatient and outpatient care (US$9.62) accounted for the majority of the direct medical costs. While patients' (US$103.77) and caregivers' productivity

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costs (US$103.77) constituted the major component of indirect cost. The intangible costs in terms of WTP value (US$266.07 vs. 88.22) and utility (EQ-5D, 0.828 vs. 0.923; QWB-SA, 0.657 vs. 0.802) were both substantially higher compared to the healthy subjects.
Conclusions: Epilepsy is a cost intensive disease in China. According to the prognostic groups, drug-resistant epilepsy generated the highest total cost whereas patients in seizure remission had the lowest cost. AED is the most costly component of direct medical cost probably due to 83% of patients being treated by new generation of AEDs.

Introduction

Epilepsy is well recognized to pose heavy economic burden on society and individual, as indicated in many cost-of-illness (COI) studies (Andlin-Sobocki et al., 2005; Begley et al., 2000; Berto et al., 2000; Bolin et al., 2012; Cockerell et al., 1994; Forsgren et al., 2005). With increasing attention towards containing health care expenditure, the economic consequences of managing epilepsy would not be spared the scrutiny. This is especially so with the advent of second-generation AEDs, the promotion of vagal nerve simulators as well as the surgical options, all of which potentially contributing to a substantial increase in the costs of managing epilepsy (Andlin-Sobocki et al., 2005; Forsgren et al., 2005). Nonetheless, it is notable that epilepsy surgery for medically intractable epilepsy in suitable candidates has consistently been found to provide favourable clinical outcomes and is associated with long-term cost savings in both adult and paediatric patient populations (Bowen et al., 2012). Besides, improved diagnostic accuracy in pre-surgical evaluation of patients is associated with reduced health resource use (O'Brien et al., 2008).

Besides increasing the utilization of health care resources, many patients with epilepsy also suffer from other co-morbidities, with mood disorders like depression being the most prominent one (Kwan et al., 2010; Vingerhoets, 2006; Wong and Lhatoo, 2000). In addition, negative psychological and social impacts are commonly detected among patients with epilepsy (Collins, 2011). These include a reduction in self-esteem, a higher probability of anxiety and unemployment/underemployment (Collings and Chappell, 1994; Jahoda, 1982), a lower marriage rate (Elliott et al., 2011; Kim et al., 2010), and difficulty with learning (Camfield et al., 2003). Moreover, the mortality of this population is higher than their healthy counterparts (Dansky et al., 1980; Smeets et al., 2007). All these factors contribute to the increase in the direct, indirect and intangible costs to patients and society.

To assess the economic burden of epilepsy, Cost-of-Illness (COI) studies are commonly employed. COI study quantifies the direct, indirect and intangible costs due to a particular disease. Generally, direct costs measure the opportunity cost of resources used for treating a particular illness, whereas indirect cost measure the value of resources lost due to a particular illness. Meanwhile, intangible cost includes pain, suffering, anxiety or fatigue because of an illness or the treatment of an illness.

To date, there were two studies estimating the economic burden of epilepsy in China (Hong et al., 2009; Liu et al., 2013), one study estimated both the direct and indirect cost (Hong et al., 2009) whereas the other one only calculated the direct cost (Liu et al., 2013). However, neither study took the caregiver's productivity cost nor the intangible cost into consideration. In cost analysis, the relevant productivity changes are those arising from both the patient and family member taking time off work in order to receive health care (Drummond et al., 2005). Thus, for a comprehensive COI study, cost of both patients' and caregiver's productivity should be included. In addition, intangible costs could be measured and valued, through ''the utility or willingness-to-pay (WTP) approach'' (Drummond et al., 2006). The inclusion of intangible cost would provide a more comprehensive estimation of the economic burden of the disease.

Hence, we aimed to estimate the direct, indirect, and intangible cost of epilepsy in China from a societal perspective. Specifically, a bottom-up, prevalence-based approach was adopted to compute the direct and indirect cost due to epilepsy, while utility and WTP values of each individual were elicited and compared with general population to describe the intangible cost. Consequently, besides providing an estimate of the direct and indirect cost of managing epilepsy, the results from our study would also provide an estimate of intangible cost of epilepsy, which is seldom reported explicitly. For informing public health, the results obtained would thus provide some important benchmark values for decision makers in assessing the economic burden of epilepsy internationally.

Methods

Subjects

The cross-sectional study recruited participants from two tertiary hospitals in Hubei Province, China: Renmin Hospital of Wuhan University, and the Fifth Hospital of Wuhan between July 2012 and January 2013. The study was approved by the Institutional Review Board of the two study sites. After informed consent was received from each participant (age > 16 years), a convenient sample of inpatients or outpatients with the diagnosis of epilepsy and a control group (without manifestation of cognitive problems, primarily from the relatives or caregivers of patients with epilepsy, hospital general staff, interns and nurses) were recruited. Attending physicians or consultant neurologists/epileptologists were responsible for initially identifying the patients. The diagnosis of epilepsy was based on clinical history, symptoms, examinations, EEG (epileptic discharges), and neuroimaging (MRI, CT) with the consensus between two physicians (SQP and LX).