Comparison of cost-of-illness with willingness-to-pay estimates to avoid shigellosis: evidence from China

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Previous studies have shown that cost of illness (COI) measures are lower than the conceptually correct willingness-to-pay (WTP) measure of the economic benefits of disease prevention. We compare COI with stated preference estimates of WTP associated with shigellosis in a rural area of China. COI data were collected through face-to-face interviews at 7 and 14 days after culture-confirmed diagnosis. WTP to avoid an episode similar to the one the respondent just experienced was elicited using a sliding-scale payment card. In contrast to previous studies’ findings, average COI estimates (2002 PPP adjusted US$28.2) approximate an upper bound estimate of WTP, rather than a lower bound. One explanation for the similarity between COI and WTP is that preventive expenditures and disutility due to pain and suffering are low for shigellosis. WTP to avoid additional cases in children aged 0–5 years is higher than in adults. Also, average COI (2002 PPP adjusted US$28.4) for children is similar to a lower bound estimate of WTP (2002 PPP adjusted US$16.4) and lies within the WTP range. Because the monetary loss associated with another episode in children is small, caregivers’ higher WTP may be attributable to the disutility of illness due to the children’s pain and suffering. These findings suggest that for some diseases, COI may approximate more comprehensive measures of economic benefits.

Keywords Cost of illness, willingness to pay, WTP, shigellosis, China
KEY MESSAGES

- For evaluating the benefits of public programmes to control shigellosis, the use of the conventional and convenient *ex post* COI figures for adults instead of *ex post* WTP measures may yield acceptable measures of the welfare impacts of reducing disease risk.

- However, the use of *ex post* COI as the estimate of the welfare impact of risk-reducing policies is likely to underestimate the benefits of programmes to prevent shigellosis in children.

- For some diseases and populations, COI may approximate the conceptually correct WTP measure of the economic benefits of disease prevention. This finding is in contrast to previous studies’ findings that COI is lower than WTP.

Introduction

Diarrhoea is a leading cause of morbidity and mortality from communicable disease in developing countries, and bacillary dysentery caused by shigellosis remains a major source of diarrhoea. The global burden of shigellosis has been estimated at 165 million cases per year, of which 163 million were in developing countries (with 1 million deaths annually) (Kotloff et al. 1999). More recent data from Asia show that the burden of shigellosis in Asia is higher than previously thought (von Seidlein et al. 2006). von Seidlein et al. (2006) also show that the burden of disease is highest among children less than 5 years of age and persons over 40 years.

In China, despite strong economic growth, bacillary dysentery remains the third most prevalent disease (Wang et al. 2004b). In 2000, 1.5 to 3.2 million episodes of bacillary dysentery occurred in China (Wang et al. 2004b). Approximately 17% of those afflicted were hospitalized. Antibiotics are frequently prescribed for bacillary dysentery in China, and overuse of antibiotics has increased multi-antibiotic resistance (Wang et al. 2004a, 2004b). The costs associated with an episode of shigellosis include out-of-pocket expenditures and lost economic opportunities, which place a burden on afflicted individuals, their families and the society at large. Ours is the first study to attempt to estimate the magnitude of the socio-economic costs associated with shigellosis in China.

A number of methodologies have been proposed for health outcome valuation, including estimation of the costs of morbidity, usually termed the cost-of-illness (COI) approach. Another, increasingly common method for evaluating the costs of morbidity and mortality, the stated preference approach, measures individual welfare change associated with a disease by directly questioning individuals in a survey, usually about their willingness to pay (WTP) for specific improvements in treatment or prevention. The COI approach is based on estimation of the direct and indirect costs associated with a disease. It may be used to estimate the ‘public’ costs to a community or society of treating infected individuals, or ‘private’ (out-of-pocket) costs to individuals, or both. The present study was concerned exclusively with private costs because COI estimates do not capture the pain and suffering, preventive expenditures associated with illness, or the value of reduced mortality risk (Harrington and Portney 1987). The few empirical studies that have compared WTP estimates with COI estimates have found that WTP is at least 1.6 to 8.0 times larger than COI (Rowe and Chestnut 1985; Chestnut et al. 1988, 1996; Dickie and Gerking 1991; Alberini and Krupnick 2000) (see Table 1). This finding is robust to whether the study focused on comparisons of *ex ante* or *ex post* measures of COI and WTP. *Ex ante* (literally ‘from beforehand’) estimates concern prospective costs/benefits of a particular action or experience, if it were to occur; *ex post* (‘hindsight’) estimates reckon costs/benefits from an action or experience that has already occurred.

The findings are also robust to variations in other study features, including study location, disease and study population. Three of these studies were conducted in the United States, one in Taiwan and one in Ethiopia. The studies measured the economic impacts of a range of acute and chronic health problems with varying severity, including minor respiratory symptoms, angina, asthma and malaria. In three of the studies, the study population was the general population, while the other two examined a population of persons with disease. Only one study was health care facility-based. Most of the studies essentially measured COI as medical expenditures net of insurance expenditures from the private perspective, rather than from the health facility, government or social perspective.

Our approach compares estimates of private COI with WTP estimates obtained from the same individuals, all of whom had recently experienced shigellosis either as an adult patient or as caregiver to a child patient. These estimates are based on data collected in the course of a combined epidemiological and economic study conducted in a rural area in Hebei Province, China, in 2002. The epidemiological component of the study was conducted through a population- and treatment-centre-based surveillance system in which the diagnosis of shigellosis in patients was confirmed by laboratory tests. In the economic study, patients with laboratory-confirmed shigellosis (or their caregivers) were asked to answer questions about their out-of-pocket expenditures and lost earnings associated with shigellosis. Respondents were also asked about their willingness to pay to avoid another episode of shigellosis like the one they had just experienced.

By comparing self-reported COI with stated preference estimates of WTP, we aimed to test three hypotheses. First, although economic theory posits that COI measures are a lower bound on WTP measures, in the present situation we expect...
COI measures to approximate WTP measures, because (1) preventive expenditures to avoid shigellosis were non-existent in the study population, (2) the disutility (pain and suffering) associated with shigellosis was not large, and (3) people did not believe the illness to be fatal. Second, several studies have found that willingness to pay for improvements in health may vary depending on whether adults or children are involved. We thus tested whether age was a significant determinant in explaining any disparity between COI and WTP measures in our survey. Third, we expected that an individual's willingness to pay to avoid another episode of shigellosis would be positively associated with the out-of-pocket expenses incurred during that individual's recent episode of the disease.

The study reported here is the first to estimate costs or benefits associated with shigellosis in China, to obtain economic data from laboratory-confirmed shigellosis patients in China, and to compare COI estimates with WTP estimates obtained in the same time period from the same individuals.

### Background

Shigellosis is a disease characterized by diarrhoea, often containing blood and mucus, accompanied by fever and abdominal pain (Keusch and Bennish 1998). Health statistics show that incidence in China in 2000 was 0.8 cases per 1000 persons, and morbidity and mortality due to shigellosis were highest in children under 5 years old (Wang et al. 2004b). An epidemiological study in 2002 found that the annual incidence rate in our study area (4.4 cases per 1000) was much higher than the national rate. Incidence was highest in children 3–4 years of age (32 cases per 1000) and in people more than 60 years of age (7 cases per 1000) (Wang et al. 2004c).

No shigellosis-related deaths were reported during the study.

Most of the diarrhoea episodes identified in our epidemiological study, including those that were ultimately diagnosed as shigellosis, received early treatment with antibiotics (e.g. oral gentamicin, norfloxacin or ampicillin), oral rehydration solution and antidiarrhoeal medications (Wang et al. 2004a). Individuals residing in the study area were within 10 minutes’ walk of a health care facility, and village health workers frequently prescribed antibiotics for diarrhoea and dysentery. More than 90% of shigellosis episodes in the study area proved resistant to antibiotics such as co-trimoxazole, ampicillin and nalidixic acid. To date, resistance by *Shigella* strains to nalidixic acid, the first-line drug recommended by WHO for the treatment of shigellosis, has rarely been observed in China (Kain et al. 1991).

Improvements in water supply, sanitation, food safety and community awareness of preventive measures are the basic public health measures used to prevent cases of diarrhoeal disease. But in areas where those basic interventions cannot be implemented in the near future, the introduction of vaccines has also been considered as an option for preventing shigellosis. Continuing efforts to develop shigellosis vaccines have seen little success because shigellosis antigens are highly serotype-specific, and there may be little or no cross-protection between species and serotypes (WHO 1997; Acosta et al. 2004). To date, the only licensed vaccine against shigellosis is the FS vaccine, which can be purchased in China (Acosta et al. 2004). It is licensed only for people aged 5 and above (Tu et al. 1999), but the major burden of *Shigella* infection is among children aged 1–4. In addition, FS is expensive, and individuals must pay for it as a non-Expanded Program of Immunization (EPI) vaccine, which results in limited use in poor, rural areas where it is most needed (DeRoeck and Nyamete 2001). (The Chinese government provides the basic EPI vaccines—BCG, OPV, DPT, measles—virtually free of charge; others, like FS, can be obtained by individuals who are willing to pay the full cost.)

### Data and methods

#### The passive surveillance study

The study area consists of 29 villages in four rural townships (Nan Gang, Nan Niu, Wu Xing and Yong An) in Zhengding County, Hebei Province, China. The total population of the four townships in 2000 was 77,121. Most residents depend on agriculture for their living.

All health care providers working in the 101 village clinics and four township hospitals in the study area were included in the shigellosis surveillance system. Between 1 January and 30 December 2002, 10,105 diarrhoea or dysentery cases were detected. For each patient, health care providers completed a case report with information on the patient’s medical history.

### Table 1 Summary of empirical analyses comparing COI to WTP measures

<table>
<thead>
<tr>
<th>Author (date), Elicitation format in CV</th>
<th>Study country</th>
<th>Disease or symptom</th>
<th>Study population</th>
<th>Approach</th>
<th>COI</th>
<th>WTP</th>
<th>WTP/COI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Alberini &amp; Krupnick (2000), Dichotomous choice format</td>
<td>Taiwan</td>
<td>Minor respiratory symptoms (e.g. cough, sore throat)</td>
<td>General population</td>
<td>ex post</td>
<td>ex post</td>
<td>1.6–2.3</td>
<td></td>
</tr>
<tr>
<td>Berger et al. (1987), Bidding game, open-ended format</td>
<td>US</td>
<td>Light symptoms (e.g. coughing, sore throat)</td>
<td>General population</td>
<td>ex post</td>
<td>ex post</td>
<td>3.1–79.0</td>
<td></td>
</tr>
<tr>
<td>Chestnut et al. (1988, 1996), Open-ended format</td>
<td>US</td>
<td>Angina</td>
<td>Health care facility based (two medical centres in California)</td>
<td>ex post</td>
<td>ex post</td>
<td>2.0–8.0</td>
<td></td>
</tr>
<tr>
<td>Cropper et al. (2004), Dichotomous choice format</td>
<td>Ethiopia</td>
<td>Malaria</td>
<td>General population</td>
<td>ex ante</td>
<td>ex ante</td>
<td>2.0</td>
<td></td>
</tr>
<tr>
<td>Rowe &amp; Chestnut (1985), Open-ended format</td>
<td>US</td>
<td>Asthma</td>
<td>People with diagnosed asthma</td>
<td>ex post</td>
<td>ex post</td>
<td>1.6–2.3</td>
<td></td>
</tr>
</tbody>
</table>
and physical examination; project staff then collected two rectal swabs or a stool specimen. The specimens were packed in a refrigerated box and delivered to a central laboratory, usually within under 4 hours of collection. Laboratory-confirmed shigellosis cases totalled 331, 3% of all diarrhoea or dysentery cases reported.

Cost-of-illness (COI) data
Of these 331 culture-confirmed shigellosis patients, 307 (202 adults and 105 primary caregivers for children aged 0–17) completed private cost-of-illness (COI) questionnaires at 7 and 14 days after the diagnosis, through face-to-face interviews. Respondents reported their households’ out-of-pocket expenditures related to treatment and recovery along with missed days of work or school due to the illness. For analysis the resulting data were categorized as reflecting direct medical, direct non-medical and indirect costs. Direct medical costs were expenditures on laboratory tests, medicines, treatment and overnight stays in health facilities. Direct non-medical costs were expenditures on transportation, lodging and meals during treatment and recovery. Indirect costs, defined as lost productivity, were calculated as the product of days of work missed by the patient and all household members and the monetary value of lost productivity, proxied by the average daily wage (for adults) in the study area. The daily productivity losses for sick children were assumed to equal 25% of the average daily adult wage for children aged 0–9 and 50% of the average daily adult wage for children aged 10–17.

Willingness-to-pay (WTP) data
We chose a stated preference method to measure WTP (contingent valuation). Revealed preference methods were not practical in this setting because there were few or no market exchanges associated with disease prevention. Vaccines against shigella were not widely used, and the costs of prevention activities for diarrhoeal disease (handwashing, water treatment, improved personal hygiene) were difficult to measure because they involve non-market activities and joint production (i.e. these activities prevent numerous diseases and also produce other valued attributes, including cleanliness).

At the end of the follow-up (14th-day) COI interview, we asked each respondent two open-ended WTP questions using a sliding-scale payment card. The respondent was asked to mark two prices on the payment card, a printed scale of prices between US$0 and US$2783. Starting at the bottom of the card (the low prices) and moving up the scale, the respondent was to mark the highest amount that he (or she) was completely sure that he would pay to avoid another episode of shigellosis like the one recently experienced. (This set a lower bound on WTP.) Then, starting at the top of the card (the high prices) and moving down the scale, the respondent was to mark the lowest amount he was completely sure that he would not pay to avoid another episode of shigellosis like the one recently experienced. (This set an upper bound on WTP.)

An open-ended elicitation format was chosen because the field conditions did not permit large enough pretests of the contingent valuation questions to support a careful selection of bid amounts for closed-ended elicitation questions. We chose the sliding-scale payment card elicitation format because, unlike a simple open-ended format that asks respondents for a point estimate, the sliding scale format measures respondents’ degree of certainty about their WTP values. While traditional payment card formats can be used to elicit a WTP range, in this payment card format respondents are asked about their upper and lower bound WTP values separately (Whittington et al. 2002). While we believe that this format encourages respondents to carefully consider both the upper and lower bounds on WTP, there have been no experiments that have compared this elicitation format with alternatives.

It is important to note that the WTP estimates from our sliding-scale questions are ex post measures. A person’s valuation of a change in health status may depend on whether the condition is described as a current or past outcome of infection, or as a potential or future outcome of infection (Johnson et al. 1998). Ex post, or certain (that is, not potential), WTP estimates require respondents to assess the prospect of expenditure on the basis of prior or present-day experience—in our case, to consider the prospects of eliminating the risk of infection, given that the respondents have had recent experience (i.e. have been infected) with the disease. Ex ante, or potential, WTP measures require respondents simply to assess future possibilities—that is (to continue our example), to consider the possible costs of becoming infected whether or not they have had any experience of the disease. When unaffected individuals are asked to evaluate prospective costs of contracting a disease, they may be uncertain about the likelihood of becoming infected and the effects of illness (on uncertainty effects see, further, Morgan and Henrion 1990). By contrast, when we asked respondents who had recently experienced shigellosis about their willingness to pay ‘to avoid another episode of shigellosis like the one they had already experienced’, they were fairly certain about the welfare losses associated with their episode of the illness, such as expenditures to treat the disease, lost productivity and disutility (pain and suffering) due to the disease. They are also more certain about the risk of illness, the severity of illness, effectiveness of prevention activities, opportunities for treatment and the effectiveness of treatment. Ex post WTP estimates are likely to differ from ex ante WTP estimates because respondents providing ex post WTP responses have less uncertainty about disease outcomes. Our valuation questions were carefully designed to ask respondents to speculate about a situation in which they would definitely become infected in the next year with an episode of shigellosis like the one they had already experienced, and then asked how much they would be willing to pay to avoid such a condition.

Comparing COI and WTP measures
COI is also an ex post estimate of the costs actually incurred by the patient (and family), made after having experienced an infection. We compared our estimates of ex post COI and ex post WTP to avoid infection using models developed by Dickie (2003), Freeman (2003) and Harrington and Portney (1987). Marginal willingness to pay for an improvement in health outcomes is typically analysed in terms of four elements: lost earnings, medical expenditures, the disutility of illness (monetized by dividing by the marginal utility of income) and preventive (also termed defensive or averting) expenditures, which are the costs of actions taken to prevent or reduce
adverse health effects (Harrington and Portney 1987). As the magnitudes of these elements vary across the sample, WTP also varies. For example, respondents with good access to low-cost health care will have lower medical expenditures than households with poor access (e.g. they must travel long distances or wait in long queues) to high-cost health care. All else equal, the respondents who have lower medical expenditures—those respondents who have good opportunities for treatment—will have a lower WTP than respondents who do not have such opportunities. Harrington and Portney (1987) and Kenkel (1994) conclude that COI is not a good approximation of WTP because COI measures only the first two elements and does not measure disutility (pain and suffering) or preventive expenditures. Policymakers nevertheless often prefer COI estimates because of their interest in the budgetary effects of health policy interventions, their ambivalence about the usefulness of economists’ welfare-theoretic estimates of economic benefits, and their scepticism about the accuracy of stated preference techniques. Our comparative analysis, based on ex post COI and ex post WTP measures, demonstrates that COI and WTP are similar for shigellosis, in some cases suggesting that either measure may be appropriate for valuation.

Results

Socio-economic and demographic characteristics of the sample respondents

Table 2 summarizes the socio-economic and demographic characteristics of the sample of 202 adult patients and 105 primary caregivers (for patients aged 0–17) who completed two survey interviews (7 and 14 days after diagnosis of shigellosis).

According to provincial data, per capita monthly income in 2003 was about US$184 (2002 PPP adjusted) in Zhengding County, or US$2218 PPP annually. Average monthly income of adult patients in our data was considerably higher: US$262 PPP, or US$3146 PPP annually. The average respondent was 34 years old. Heads of household were predominantly farmers (71%). Of adult patients overall, 33% were wage workers. Most patients (99%) lived in owner-occupied housing, and average (self-reported) market value of these homes was US$17,416 PPP.

The average length of a shigellosis episode was 4.5 days. More than half of working-age adult patients (105 of 202) lost 2 days’ wages due to shigellosis, and about the same proportion of children aged 6–17 years (31 of 55) missed 2 days of school. Most patients did not have substitute labourers to replace their work activities while they were sick. Household members did not report missing any work while taking care of patients at home or while accompanying patients to health care providers for treatment and recovery.

We also inquired about personal and household attributes and preventive behaviours that might reduce the risk of infection with shigellosis. A large majority of patients (more than 70%) reported that they always washed their hands with soap and use boiled water for drinking and cooking. About 50% of respondents reported that they consumed ‘street food’ (from vendors), which is presumed more likely to be contaminated than home cooking, more than three times a week.

<table>
<thead>
<tr>
<th>Variable</th>
<th>Mean</th>
<th>S.D.</th>
</tr>
</thead>
<tbody>
<tr>
<td>General</td>
<td>77 121</td>
<td>2218</td>
</tr>
<tr>
<td>Age (continuous)</td>
<td>33 8</td>
<td>23 5</td>
</tr>
<tr>
<td>Age categories (dummy)</td>
<td>0 0</td>
<td>0 2</td>
</tr>
<tr>
<td>Age 0–1</td>
<td>0 1</td>
<td>0 3</td>
</tr>
<tr>
<td>Age &gt;17</td>
<td>0 7</td>
<td>0 5</td>
</tr>
<tr>
<td>Female (dummy)</td>
<td>0 5</td>
<td>0 5</td>
</tr>
<tr>
<td>Daily income if adults (US$ PPP)</td>
<td>12 0</td>
<td>19 8</td>
</tr>
<tr>
<td>Daily income if adults (US$ PPP)</td>
<td>15 2</td>
<td>13 8</td>
</tr>
<tr>
<td>Daily income if adults (US$ PPP)</td>
<td>3 2</td>
<td>3 2</td>
</tr>
<tr>
<td>Daily income if adults (US$ PPP)</td>
<td>0 5</td>
<td>0 9</td>
</tr>
<tr>
<td>Daily income if adults (US$ PPP)</td>
<td>23 0</td>
<td>52 4</td>
</tr>
<tr>
<td>Daily income if adults (US$ PPP)</td>
<td>17 5</td>
<td>11 0</td>
</tr>
<tr>
<td>Daily income if adults (US$ PPP)</td>
<td>0 9</td>
<td>0 3</td>
</tr>
</tbody>
</table>

Univariate analysis

COI measures

Table 3 presents the components of ex post private COI per episode for children and for adults using the purchasing power parity (PPP) exchange rates used in this analysis are available from the IMF website: http://imf.org/external/pubs/ft/weo/2004/02/data/dbgim.cfm. Using PPP US dollar exchange rate, US$1 = Chinese Yuan 1.8 in 2002. We assume US$1 = Chinese Yuan 8.3. Therefore, US$1 = PPP adjusted US$4.6 in 2002, China.

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The total COI for children and for adults is not significantly different ($P = 0.15$).

Direct medical costs per episode were US$25.7 PPP for children and US$17.9 PPP for adults. Direct COI accounted for 97% of total private COI for children; direct non-medical and indirect costs accounted for only 3% of total private costs. As shown in Figure 1, direct COI and total COI for children have similar distributions, probably because 60% of episodes in child patients did not incur indirect COI from their episodes of shigellosis. A large proportion (70%) of children’s caregivers paid less than US$23.0 PPP, and only 10% paid more than US$69.0 PPP in out-of-pocket expenditures associated with their illness. Among adults, direct medical costs (US$18.0 PPP, 60% of total COI) and the patient’s lost productivity (US$10.1 PPP, 38% of total COI) were the major components of the ex post private COI. Figure 2 shows that 50% of adults paid less than US$11.5 PPP and only 2.5% paid more than US$64.0 PPP in out-of-pocket expenditures. About one-third of adult patients did not lose any wages or productivity due to their illness, and 70% lost less than US$11.5 PPP. Total private COI due to shigellosis was less than US$34.5 PPP for 80% of adult patients.

Among all 307 patients, only three children and one adult patient were hospitalized and had much higher ex post COI (US$381.3 PPP) compared with non-hospitalized cases (US$23.0 PPP). Average COI for non-hospitalized cases is significantly different from COI for hospitalized cases at the $P = 0.0001$ level ($z = -3.31$).

Table 4 shows total private COI by age group using PPP exchange rates. Average COI for children aged 2–5 years (US$31.1 PPP) was highest, followed by adults (US$28.1 PPP), older children (6–17 years, US$27.5 PPP), and infants (0–1 years, US$22.0 PPP). The total range in average costs was US$9.2 PPP, representing a 42% difference between the lowest to highest. Nevertheless, the rank-sum tests indicate no significant difference between any pair of two age groups ($P = 0.01, z = -0.40$).

**Stated preference (WTP) measures**

When replying to our sliding-scale payment card WTP questions, 11 respondents (3.9%) indicated that they would not want to pay anything to avoid a future episode of shigellosis as severe as the one just suffered. For all ages taken together, the average lower bound of WTP estimates was US$12.8 PPP,

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### Table 3

<table>
<thead>
<tr>
<th>Components of COI</th>
<th>Children ($n = 105$)</th>
<th>Adults ($n = 202$)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Direct cost of illness</td>
<td>26.5 (49.5)</td>
<td>18.0 (59.6)</td>
</tr>
<tr>
<td>Medical cost</td>
<td>25.7 (44.0)</td>
<td>17.9 (58.5)</td>
</tr>
<tr>
<td>Non-medical cost</td>
<td>0.7 (6.6)</td>
<td>0.1 (1.3)</td>
</tr>
<tr>
<td>Indirect cost of illness</td>
<td>1.9 (3.2)</td>
<td>10.1 (13.8)</td>
</tr>
<tr>
<td>Total cost of illness</td>
<td>28.4 (50.2)</td>
<td>28.1 (65.5)</td>
</tr>
</tbody>
</table>

### Figure 1

Cumulative distribution of total estimated private COI, direct COI and indirect COI due to an episode of shigellosis in children aged 0–17 years. For the purpose of presentation, this figure excludes one observation with extremely high total COI and direct COI (> US$120 PPP).

### Table 4

<table>
<thead>
<tr>
<th>Age (years) group</th>
<th>Annual incidence rate</th>
<th>Lower bound</th>
<th>Upper bound</th>
</tr>
</thead>
<tbody>
<tr>
<td>0–1 ($n = 12$)</td>
<td>14.0</td>
<td>22.0 (35.0)</td>
<td>16.7 (9.2)</td>
</tr>
<tr>
<td>2–5 ($n = 43$)</td>
<td>19.0</td>
<td>31.1 (71.1)</td>
<td>23.1 (84.0)</td>
</tr>
<tr>
<td>6–17 ($n = 50$)</td>
<td>3.3</td>
<td>27.5 (26.8)</td>
<td>10.6 (13.6)</td>
</tr>
<tr>
<td>0–17 (children) ($n = 105$)</td>
<td>7.9</td>
<td>28.4 (50.2)</td>
<td>16.4 (54.6)</td>
</tr>
<tr>
<td>&gt;17 (adults) ($n = 202$)</td>
<td>4.8</td>
<td>28.1 (65.5)</td>
<td>11.0 (35.7)</td>
</tr>
<tr>
<td>All ages ($N = 307$)</td>
<td>4.4</td>
<td>28.2 (60.6)</td>
<td>12.8 (43.1)</td>
</tr>
</tbody>
</table>

*Unit of annual incidence rate is per 1000 people per year.

**Figure 2** Cumulative distribution of total estimated private COI, direct COI and indirect COI due to an episode of shigellosis in adults (ages 18+). For the purpose of presentation, this figure excludes one observation with extremely high total COI and direct COI (> US$120 PPP).

**Stated preference (WTP) measures**

When replying to our sliding-scale payment card WTP questions, 11 respondents (3.9%) indicated that they would not want to pay anything to avoid a future episode of shigellosis as severe as the one just suffered. For all ages taken together, the average lower bound of WTP estimates was US$12.8 PPP,
and the average upper bound was US$28.0 PPP (see Table 4). Estimates were compared using the Wilcoxon rank-sum test, as data were not normally distributed. Average lower bound for children (US$16.4 PPP) was slightly higher than that for adults (US$11.0 PPP) \((P = 0.005)\). But average upper bound was much higher for children (US$38.7 PPP) than for adults (US$22.5 PPP) \((P = 0.002)\). Average lower bound of WTP was highest for children aged 2–5 (US$23.1 PPP), followed by children aged 0–1 (US$16.7 PPP); the lesser values, for adults (US$11.0 PPP) and older children (6–17 years, US$10.6 PPP), were nearly the same. Average upper bound WTP was 2 to 2.5 times larger than average lower bound by age group. Average upper bound was highest for children aged 2–5 (US$51.9 PPP), followed by children aged 0–1 (US$38.9 PPP), children aged 6–17 (US$27.3 PPP), and adults (US$22.5 PPP). The highest average upper bound (US$51.9 PPP, for children 2–5) was four times larger than the lowest average lower bound (US$10.6 PPP, for children 6–17). Caregivers for children aged 2–5 years had the highest lower and upper bound WTP, perhaps because they recognized from personal experience or from acquired knowledge that children were most vulnerable to shigellosis infection. (Children aged 3–4 had the highest incidence rate.)

Figure 3 shows the degree of the certainty for respondents’ WTP estimates. Estimates are ordered from left to right by lower bound WTP, from highest to lowest value. There is a vertical line for each respondent and it represents their WTP range, from their lower bound to their upper bound. A longer vertical line indicates a larger interval between lower and upper bound and greater uncertainty, while a shorter vertical line indicates less uncertainty. Eighteen per cent of responses showed a relatively small interval between lower and upper bound, less than US$5, indicating a relatively small range of uncertainty. Figure 3 shows that respondents with higher lower bounds (toward the left side of the graph) tended to report more uncertainty (larger WTP ranges) than other respondents. That is, respondents who reported that they were willing to pay a higher amount for the lower bound tended to have the greatest uncertainty about their WTP. This finding is similar to Whittington et al.’s (2002) findings regarding WTP for HIV/AIDS vaccines in Guadalajara, Mexico. After five extreme values for lower/upper bounds were dropped to control their impact on the mean value, the average range between lower and upper bound WTP estimates came to US$12.0 PPP.

**Comparison of COI and WTP measures**

For our entire sample (adult patients and child patients as represented by their adult caregivers), the average self-estimated private COI of a recent episode of shigellosis was US$28.2 PPP. The average upper bound of WTP measures for the entire sample was US$28.0 PPP. A rank-sum test indicates that this small apparent difference is not significant \((P = 0.30)\). But the average lower bound of WTP (US$12.8 PPP) is significantly lower than average COI \((P = 0.0001, z = 11.28)\). These results show ex post COI estimates that approximate the upper bound of ex post WTP, not the lower bound.\(^2\) This finding is not consistent either with a general assumption in health economics that COI is a lower bound of WTP, or with several other empirical analyses comparing ex post COI with ex post WTP estimates that have reported WTP/COI ratios ranging from 1.6 to 79.0 (see Table 1).

Figure 4 compares lower and upper bounds of WTP to COI by age group. The lower bound of WTP for episodes in children aged 0–5 is similar to the COI measure, because the upper bound WTP estimate was much higher than that for older children and adults. COI estimates for people more than 5 years old are similar to upper bounds of WTP estimates, except for the elderly (older than 60 years). The COI for the elderly was much higher than the upper bound of WTP.

Further, when caregivers of child patients are subcategorized as parents or non-parents, lower (US$29.4 PPP) and upper bounds (US$68.1 PPP) of WTP for children whose primary caregivers were parents were approximately three times greater than those for children whose primary caregivers were non-parents (US$9.2 PPP, US$23.0 PPP) \((P = 0.006)\). COI for children whose caregivers were their parents was not significantly different than COI for children with non-parental caregivers \((P = 0.70)\). We interpret these results to mean that the disutility to parents caused by their children’s shigellosis was much higher, and that altruistic willingness to pay to protect their own children from the disease may be one of the
main reasons for the great disparity between COI and WTP estimates. We speculate that this result may be widespread in China because of the nation’s ‘one child’ policy: parents might have higher disutility from sickness in an only child.

WTP and COI estimates also vary by township (Table 5). For children, COI estimates from respondents living in Nan Gang, the poorest town in the study area, were lowest (US$13.3 PPP). Lower and upper bounds of WTP estimates from Nan Gang (US$3.2 PPP, US$10.6 PPP) were lowest, too. Lower and upper bounds of WTP from respondents in Yong An, the richest township, were the second lowest. Residents of Wu Xing had the highest COI (US$40.0 PPP), perhaps explained by their high expressed preference (100%) for the village clinic as a treatment option for diarrhoea. Their lower and upper bounds of WTP were higher (US$17.5 PPP, US$35.0 PPP), as well.

### Multivariate analyses

We conducted multivariate analyses to further investigate the determinants of the COI estimates and the WTP range. While the COI data are comprised of points from a continuous distribution, the WTP data are interval data representing the lower and upper bounds on true WTP. We used least squares regression to analyse COI and interval regression to analyse the WTP range. We estimated models with normal and log-normal distributions and selected the log-normal model because it had the better goodness-of-fit. We use a robust interval regression estimator to adjust for heteroscedasticity.

#### Choice of variables for the multivariate analysis

Independent variables in the multivariate analyses include age of the patient, gender of the respondent, township of residence, whether the adult respondents worked for wages, and weekly food expenditures. Age is generally found to be the most consistent predicator of WTP and COI measures.

We omitted income from our model specifications owing to a lack of good income measures for our sample. Proxy income variables such as expenditure on electricity and water were not appropriate because government subsidies for these commodities distort the relationship between such expenditures and income. Because it was not subsidized by the government, expenditure on food or health care was an acceptable proxy for income, but our data on respondents’ health care expenditures during the week before they presented at health care facilities was strongly correlated with COI measures. Ultimately we retained respondents’ status as wage-earner (yes/no) as an indicator of higher income levels.

To test whether a respondent’s WTP to avoid another episode of illness is related to out-of-pocket expenditures due to the illness, our interval regression models of the determinants of the WTP range include direct COI. We would expect to see a positive relationship between WTP and direct COI.

Costs of preventive behaviour are important components in explaining disparity between COI measures and WTP measures (Harrington et al. 1989; Dickie and Gerking 1991; Dickie 2003). However, it is not clear just how to measure the cost(s) of averting a single episode of shigellosis. A list of such preventive activities might include personal hygiene (careful handwashing after using the toilet, or washing hands with soap), safe food preparation, boiling drinking and cooking water, protecting sources of food and water from contamination, and practising hygienic disposal of household wastes. To take handwashing as an example, preventive expenditures would include the cost of obtaining soap, the cost of water (if any), and the cost of time spent washing hands. Computing such expenditures is problematic because preventive actions produce joint products (see Dickie and Gerking 1991). In our case, for instance, hand-washing reduces the risk not only of shigellosis, but also of other diseases. Moreover, the number of times the hands must be washed to prevent a single case of shigellosis is unknown. For these reasons we shifted focus slightly and examined how preventive behaviours (rather than preventive expenditures) affected COI and WTP estimates. To measure preventive behaviours, we asked the respondent to report the regularity with which s/he: washed hands after visiting the toilet, boiled drinking and cooking water, and avoided the consumption of food purchased from street vendors.

#### Determinants of COI and lower bound of WTP estimates

Table 6 presents results from our multivariate models. The dependent variable for the first three specifications is the total private COI estimate. The dependent variable for the other three specifications is the interval created by the lower bound and upper bound of the WTP range. Individual characteristics such as age, township of residence, gender and weekly food expenditures are included in all six models. Models 4 to 6, the WTP models, also include variables indicating whether the respondent worked for a wage, preventive behaviours and the number of sick days. These variables are excluded from the COI specifications because (1) lost wages and number of sick days are components of COI; and (2) more frequent preventive behaviours can reduce the risk, but probably not the severity, of diarrhoeal disease, whereas COI measures may have a positive relation to severity of shigellosis.

Among models based on all cases (1 and 4), only the variable ages 0–1 is significant in the COI model (model 1), but in the WTP model (model 4), ages 0–1, ages 2–5 and ages 6–17 are significant. Respondents who lived in Nan Gang had lower COI and WTP measures we hypothesize that income affects WTP, and some regions have higher incomes than others, so
ceterus paribus, regions should matter. Respondents who lived in Nan Niu also had lower COI, but they expressed higher WTP estimates than those living in the other townships, consistent with the results of the univariate analysis. Respondents' gender was not a statistically significant factor in the WTP models. Other things being equal, most recent weekly food expenditure was not significant in WTP models, but in COI models it is associated with increases in total COI.

In the interval regression analysis of WTP (model 4), the coefficient on the direct COI variable in the WTP models indicates that WTP increases with direct COI, but decreases with indirect COI. Since total COI for all ages is comprised of 74% direct COI and 26% indirect COI, on average, the average net effect of total COI on WTP is positive.

The indicators of preventive behaviour—frequency of consumption of street food and hand-washing—were not a significant determinant of the WTP measures, except for adults (model 5). The finding that hand-washing was not a significant variable was unexpected, as several studies have demonstrated that improvements in hand-washing practices can lead to an appreciable reduction (14–40%) in morbidity caused by diarrhoeal disease (Black et al. 1981; Rahman 1983; Kaltenhaler et al. 1988; Pinfold et al. 1988; Stanton and Clemens 1988; Wilson et al. 1991). Several explanations can be offered for this lack of a relationship between hand-washing and WTP estimates in our data: (1) respondents might have attributed recent experience with diarrhoea to other causes; (2) respondents might have adopted preventive behaviours for general hygienic purposes, not simply to avoid only one episode of shigellosis; and/or (3) respondents were asked only about the frequency of hand-washing with soap, whereas the important criterion may be adequacy of the procedure, not frequency. Hoque et al. (1995a,b) found that most respondents (in a survey in Bangladesh) said they washed their hands after defecation, but the hand-washing methods varied. Hoque further reported that after post-defecation hand-washing, the hands remained highly contaminated when washing procedures were not adequate (Hoque 2003). (He defines appropriate procedure as use of both hands, use of a cleansing agent, rubbing hands with the agent, rinsing with water, and drying.)

### Table 6: Determinants of COI and WTP interval (t-statistics in parentheses)

<table>
<thead>
<tr>
<th>Variables</th>
<th>COI&lt;sup&gt;a&lt;/sup&gt;</th>
<th>WTP interval&lt;sup&gt;b&lt;/sup&gt;</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Model 1 All</td>
<td>Model 2 Adult</td>
</tr>
<tr>
<td>Intercept</td>
<td>1.65*** (17.15)</td>
<td>1.85*** (12.97)</td>
</tr>
<tr>
<td>Direct COI</td>
<td>0.01*** (4.74)</td>
<td>0.01*** (5.79)</td>
</tr>
<tr>
<td>Indirect COI</td>
<td>−0.02** (−2.03)</td>
<td>−0.02* (−1.79)</td>
</tr>
<tr>
<td><strong>Socio-economic</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age 0–1</td>
<td>−0.51** (−2.33)</td>
<td>−0.62** (−2.33)</td>
</tr>
<tr>
<td>Age 2–5</td>
<td>−0.20 (−1.64)</td>
<td>−0.23 (−1.42)</td>
</tr>
<tr>
<td>Age 6–17</td>
<td>0.04 (0.32)</td>
<td></td>
</tr>
<tr>
<td>Age 18–29</td>
<td>−0.32** (−2.04)</td>
<td></td>
</tr>
<tr>
<td>Age 30–39</td>
<td>−0.19 (−1.20)</td>
<td>0.01 (0.10)</td>
</tr>
<tr>
<td>Age 50–59</td>
<td>−0.07 (−0.46)</td>
<td>0.10 (0.61)</td>
</tr>
<tr>
<td>Age 60+</td>
<td>−0.12 (−0.89)</td>
<td>−0.04 (−0.37)</td>
</tr>
<tr>
<td>Female</td>
<td>0.001 (0.01)</td>
<td>0.05 (0.46)</td>
</tr>
<tr>
<td>Food expenditures (per week)</td>
<td>0.06*** (3.89)</td>
<td>0.05** (2.01)</td>
</tr>
<tr>
<td>Wage worker</td>
<td>0.31*** (3.73)</td>
<td>0.31*** (3.52)</td>
</tr>
<tr>
<td><strong>Townships</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nan Gang</td>
<td>−0.53*** (−5.01)</td>
<td>−0.59*** (−4.76)</td>
</tr>
<tr>
<td>Nan Niu</td>
<td>−0.49*** (−4.47)</td>
<td>−0.61*** (−4.35)</td>
</tr>
<tr>
<td>Yong an</td>
<td>0.28* (1.82)</td>
<td>0.26 (1.48)</td>
</tr>
<tr>
<td><strong>Preventive behaviour</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Buying street food (1 = &gt;3 times/week)</td>
<td>0.09 (1.13)</td>
<td>−0.01 (−0.09)</td>
</tr>
<tr>
<td>Washing hands (1 = &gt;50% of the time)</td>
<td>−0.03 (−0.31)</td>
<td>0.14* (1.86)</td>
</tr>
<tr>
<td>No. of sick days</td>
<td>0.01 (0.31)</td>
<td>0.01 (0.57)</td>
</tr>
<tr>
<td>R²</td>
<td>0.20</td>
<td>0.23</td>
</tr>
<tr>
<td>Wald χ²</td>
<td>305.76</td>
<td>274.19</td>
</tr>
<tr>
<td>Log pseudo-likelihood</td>
<td>−442.53</td>
<td>−282.63</td>
</tr>
<tr>
<td>N</td>
<td>307</td>
<td>202</td>
</tr>
</tbody>
</table>

<sup>a</sup>P < 0.10; **P < 0.05; ***P < 0.01.

These results are based on a least squares regression model with a log-normal error distribution.

These results are based on an interval regression model with a log-normal error distribution.
Models 5 and 6 were estimated separately for adults and for children (aged 0–17). WTP estimates for adult patients increased with direct COI and decreased with indirect COI (model 5). Since adult COI is comprised of 60% direct COI and 38% indirect COI, on average, the average net effect of total COI on WTP for adults is negative. Children’s caregivers’ WTP estimates are also positively affected by direct COI (model 6), but the statistical significance of this parameter is lower than in the adult WTP model. The WTP estimates for children are not affected by indirect COI. These results suggest that when adult patients answered WTP to avoid another episode of illness for themselves, they considered the prospective monetary loss, such as medical expenditure and the indirect losses. But when adult caregivers estimated the value of avoiding another, similar episode of shigellosis in their children, direct costs of medical treatment were a weaker influence and the indirect COI were not considered.

The number of sick days, which is one indicator of severity, is not significant in any of the models. For the child patients (model 6), the length of illness variable was dropped because it was found to be highly and significantly correlated with the direct COI.

The regression results suggest that COI and WTP for adult patients are similar, which is consistent with the univariate analysis showing that WTP estimates associated with shigellosis are close to COI estimates. A few empirical studies that regressed WTP on COI have reported parameters estimates of −0.04 for the natural logarithmic value of out-of-pocket medical expenses (Rowe and Chestnut 1985), 0.02 for household direct COI (Cropper et al. 2004), and 3.13 for out-of-pocket expenses plus income loss (Chestnut et al. 1988, 1996).

Conclusion and discussion

This study analyses ex post private COI and ex post WTP estimates associated with shigellosis, elicited from laboratory-confirmed shigellosis patients in a rural area of Zhengding County, China, in 2002. Respondents (adult patients and caregivers of child patients) were asked to estimate private COI for the recent infection, and also to estimate WTP to avoid another episode of illness as severe as the recent one. Average self-assessed COI due to patients’ recent episodes of shigellosis was US$28.2 PPP; average COI for all children (ages 0–17) (US$28.4 PPP) was not significantly different from average COI for all adults (US$28.1 PPP). The average lower bound of WTP estimates was US$12.8 PPP, and the average upper bound was US$28.0 PPP. Average monthly per capita income in Zhengding at the time of the survey was US$184; respondents thus lost an average of 15% of their monthly income due to their recent episodes of illness. They would be willing to pay from 7% to 15% of their monthly income to avoid the certain prospect of having another episode of shigellosis as severe as the one just experienced. That is, on average, patients and adult caregivers for children were willing to pay, at a maximum, about the same amount of money to prevent another episode as they had paid to cope with a recent bout of shigellosis. Thus, the common perception that stated preference methods grossly overstate what people are willing to pay is not supported by this analysis.

We see that caregivers’ willingness to pay to avoid another episode of shigellosis for child patients was 1.5 to 1.7 times higher than WTP estimates for adult patients. Multivariate analysis showed that when adult patients estimated WTP for themselves, they apparently primarily considered both the direct and indirect costs that they had experienced with their recent episode of illness. Estimates from caregivers indicate that direct costs were a weaker influence and indirect costs were not a primary concern when they contemplated another shigellosis infection in child patients who had recently had the disease.

According to Harrington and Portney (1987), WTP to improve health is equivalent to the sum of four elements: medical expenses, lost wages, preventive expenditures, and the monetized value of the disutility of illness. COI measures are only the sum of medical expenses and lost wages. In our sample, we found that preventive expenditures do not exist or are immeasurable. Thus the monetized disutility of the disease (in our case, shigellosis) is likely the primary influence in the gap between COI and WTP. Since the mortality risk associated with shigellosis in our study population is low, we do not believe that respondents’ benefits of mortality risk reduction are significant. Consequently, COI and WTP are very similar in this sample for this disease. This finding supports the concept validity of our WTP estimates and is consistent with Pack et al.’s (2006) findings from socio-cultural studies of willingness to be vaccinated against shigellosis that respondents from households that have experienced an episode of shigellosis are less likely to be willing to receive vaccinations.

A second finding of interest from our ex post analysis was that although for children aged 0–5 years, the lower bound of WTP estimates was similar to COI, the COI estimates for patients aged 6 years and older were close to the upper bounds of their WTP estimates—a result inconsistent with theoretical expectations and published analyses of similar work.

Thirdly, we note that WTP for adults was influenced by COI, but the influence was weaker for children. We conclude that because for adults the disutility associated with one episode of shigellosis is not large, their estimated present COI is close to their upper bound of WTP for avoiding another, equally severe episode of the illness. But for children the disutility due to the disease is large enough to make their caregivers’ COI estimates equal to the lower bound of their WTP, probably because parents cared a great deal about the pain and suffering their children had experienced and would not wish them to endure future infections of the same severity.

There are several policy implications of our work. First, for evaluating the benefits of public programmes to control shigellosis, the use of the conventional and convenient ex post COI figures for adults instead of ex post WTP measures may yield acceptable measures of the welfare impacts of reducing disease risk. Second, the use of ex post COI as the estimate of the welfare impact of risk-reducing policies is likely to underestimate the benefits of programmes to prevent shigellosis in children. Third, these results could be used to inform the design of effective and efficient disease control programmes. As discussed in the introduction, the FS vaccine against shigella is expensive, which limits its use among the rural poor. While lowering the price through government subsidies or other means is one option for increasing uptake, our findings suggest...
that lower subsidies might be needed to achieve target coverage rates among children than among adults because caregivers have a strong desire to protect their children’s health.

Our WTP measures for avoiding another episode of shigellosis have limitations for generalization, as we interviewed only subjects who had recently suffered an episode of the illness. Because the study area was rural and poor, and poor households may have been more likely to acquire shigellosis, our samples respondents’ WTP estimates might be lower than what would be expected among the general population. On the other hand, because respondents in our sample had recently experienced shigellosis, their willingness to pay might be much higher than among the general population, who lacked immediate experience of the disease.

Acknowledgements
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Endnotes
1 The prices appeared in Chinese yuan in the survey (see Appendix). PPP exchange rates are used to convert yuan to the amount of 2002 US dollars that can purchase the same quantity of goods and services. These purchasing power parity (PPP) measures are adjusted for both the differences in price levels between countries and inflation. The market exchange rate in 2002 was US$1 = 8.3 yuan. The ratio of price levels in the US to price levels in China is 1.8, thus the 2002 PPP exchange rate is US$1 = 4.6 yuan. For readers’ convenience, we express US dollars in terms of PPP adjusted dollar values throughout the paper.

2 Respondents were more likely to indicate that their lower bound WTP was less than the COI if (1) they were 30–49 years old, (2) they were older than 5 years, or (3) they had been sick longer and paid higher out-of-pocket expenditures. (The results of probit regression analyses of whether the lower bound of WTP was less than the COI will be provided upon request by the authors.) The last finding deserves some discussion. China’s health care system has changed dramatically as China’s economic system has changed. Although the income gap between urban and rural households. The self-paid share in total health expenses was much greater in rural (87%) than in urban areas (44%) in 1998 (Zhang and Kanbur 2003). The greater share of the financing burden falls on poor people in rural areas of China (van Doorslaer and Wagstaff 1993). Since our study area was a poor rural area, respondents who had greater out-of-pocket expenses may have lower ex post WTP due to budget constraints.

References
Kotloff KL, Winickoff JP, Ivanoff B et al. 1999. Global burden of shigellosis infections: Implications for vaccine development and...


Appendix

Contingent valuation question

I am very sorry that you (patient’s name1) recently experienced this episode of shigellosis. Now I want to ask you a question about how much it would be worth to you to avoid getting another episode of shigellosis like the one you (patient’s name) recently experienced. In other words, I want you to imagine that it was certain that you (patient’s name) would have another episode, and then tell me how much it is worth to you for this NOT to happen. There are no right or wrong answers. I really want to know what you think.

...I want to ask you some questions about how much you would be willing to pay for you personally (patient’s name) to not have another episode of shigellosis...

Please consider the list on the following page with a range of monetary values (prices). As you know, prices affect the decisions we make. If the price of something is very high, it is very unlikely that we buy it. By contrast, if the price is very low, it is much more likely that we will buy it. We are typically less certain (more unsure) of what to do when the price is neither very high nor very low. Suppose the attached list presents a range of possible prices to avoid you having another episode of shigellosis like the episode you just had—between zero and Yuan 5000.

First, starting at the bottom of the page (the low prices) and moving up, I want you to mark the highest amount that you are completely sure that you would pay to avoid another episode of shigellosis. (Remember, we are talking now about your decision to pay for yourself (patient’s name).)

Second, starting at the top of the page (the high prices) and moving down, I want you to mark the lowest amount you are completely sure that you would pay to avoid another episode of shigellosis like the one you (patient’s name) recently experienced.

¥5000
¥4000
¥3000
¥2000
¥1000
¥900
¥800
¥700
¥600
¥500
¥400
¥300
¥200
¥100
¥90
¥80
¥70
¥60
¥50
¥40
¥30
¥20
¥10
¥5
¥1

---

1When the respondent was the shigellosis patient, they were asked about their willingness to pay to prevent another episode in themselves. When the respondent was a caregiver for a child patient, they were asked about their willingness to pay to prevent another episode in the child patient.