USE OF DISABILITY ADJUSTED LIFE YEARS IN THE ESTIMATION OF THE DISEASE BURDEN OF ECHINOCOCCOsis FOR A HIGH ENDEMIC REGION OF THE TIBETAN PLATEAU

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Abstract. Shiqu County, located on the Tibetan plateau of western China, has an extremely high prevalence of both human alveolar echinococcosis (AE), and cystic echinococcosis (CE). The short form 12 version 2 quality of life survey, which was used to evaluate the extent to which morbidity associated with echinococcosis should be accounted, verified that there was a significant reduction in the mean health scores in all categories for individuals diagnosed with abdominal echinococcosis compared with an age and sex cross-matched population. Results of a larger ultrasound survey, which screened 3,135 subjects, demonstrated that the prevalence rates of AE and CE were both approximately 6% with a combined prevalence rate of 11.4%. Prevalence rates adjusted for the age and sex structure of Shiqu County were 4.6% for AE and 4.9% for CE with an estimated overall adjusted prevalence rate of 9.5%. The burden of disease associated with echinococcosis was calculated using disability adjusted life years (DALYs) based on these estimated prevalence rates. Monte-Carlo techniques were used to model the uncertainty in the prevalence estimates and the disability weights. Using these methods, we estimated that the total numbers of DALYs lost due echinococcosis was 50,933 (95% confidence interval [CI] = 41,995–61,026). The DALYs lost consisted of approximately 32,978 (95% CI = 25,019–42,422) due to AE and 17,955 (95% CI = 14,268–22,128) due to CE and suggests an average of approximately 0.81 DALY lost per person. This study has clearly shown that the impact of DALYs lost due to echinococcosis, in terms of medical treatment costs, lost income, and physical and social suffering, is likely to be substantial in this highly endemic region of China.

INTRODUCTION

Human cystic echinococcosis (CE) and alveolar echinococcosis (AE) are caused by the larval stage of the taeniid tapeworms Echinococcus granulosus and E. multilocularis, respectively, and are among the most deadly helminth diseases known to humans. Cystic echinococcosis produces space-occupying lesions, usually in the liver or lungs, whereas AE results in highly infiltrative lesions of the liver and may give rise to metastases. Expenses and loss of health and vitality associated with Echinococcus infection can become a significant burden not only for the affected individual and his or her family, but also for the community as a whole. The Tibetan plateau region of western China has been found to have one of the highest prevalences of both human CE and AE in the world. Potential impact of the disease on afflicted individuals must be taken into consideration when constructing a disability adjusted life year (DALY) estimate. A health survey is a useful tool with which to evaluate the physical and mental health state of a person with, in this instance, echinococcosis compared with a control population. Two previous studies suggested that patients surgically treated for CE had a significant decrease in the quality of life. Subjects presenting for treatment have also been reported as having a substantially higher rate of unemployment. However, to evaluate the societal burden of disease it is important to understand the effect that CE and AE have on previously undiagnosed individuals. The short-form 12 (SF-12) version 2 health survey is a generic measure of general health and well-being that can be used to evaluate the extent to which morbidity, associated with echinococcosis, should be accounted. Therefore, the quality of life of individuals who were found to be abdominal ultrasound positive for either AE or CE on a cross-sectional study of the population of Shiqu County (Sichuan Province, People’s Republic of China) was compared with negative individuals using this instrument. It is essential to know such information about the morbidity effects of echinococcosis before the numbers of DALYs lost due to the disease can be estimated.

The DALYs were first constructed for the Global Burden of Disease Study, which was developed to attempt to quantify the worldwide burden of disease attributed to 107 causes by sex and age. This technique considers the impact of both premature mortality and morbidity caused by a disease state and can then be used to evaluate the economic impact of the disease on the community as well as the potential cost-effectiveness of intervention strategies. Human echinococcosis was not evaluated in the Global Burden of Disease Study. Therefore, DALYs have been constructed for both AE and CE and applied to a region of the Tibetan plateau (Shiqu County, Sichuan Province).

MATERIALS AND METHODS

The SF-12 version 2 health survey. The SF-12 version 2 health survey (QualityMetric, Inc., Lincoln, RI) was used in this study due to its brevity and ease of use. Eight domains, or scales, of health are assessed in the survey: physical functioning, role-physical, bodily pain, general health, vitality, social functioning, role-emotional, and mental health. These domains were chosen from among 40 recommended in the Medical Outcomes Study and are considered among the most frequently measured health concepts. In addition, two component scores, the Physical Component Summary (PCS) and the Mental Component Summary (MCS) were evaluated. The translation of the American English version of the SF-12 version 2 into Tibetan was undertaken according to the International Quality of Life Assessment protocol, which involved forward and backward translation and testing on a small pilot study. In addition, appropriate wording substitutions were made that embodied similar concepts and health
requirement levels, but were more familiar to the survey subjects.6 Scoring of the SF-12 version 2 health survey was undertaken in accordance with standard procedures.6

Subjects. From 2001 to 2003, 3,135 subjects were examined using abdominal ultrasound as part of an echinococcosis screening and epidemiologic survey for Shiqu County, which has an estimated population of 63,000.5 Prevalence estimates and an age profile of the screened population, were calculated from the results of the ultrasound survey. The age profile of the screened population was then compared with the most recent census of the population of Shiqu County and an adjusted number of cases, expected from the 3,135 subjects if they had the same age profile as the total population, was calculated.5 The adjusted prevalence for echinococcosis was then determined accordingly. Consent was obtained from all participants and individuals shown to be echinococcosis positive, based on World Health Organization diagnostic criteria, were provided free of charge with albendazole tablets as well as informed of their surgical options.14,15 Ethical approval for all work carried out in China was obtained from the Medical Sciences Expert Consultant Committee, Sichuan Provincial Health Bureau, Sichuan Province (People’s Republic of China). During April 2003, the Tibetan version of the SF-12 version 2 health survey was administered to ultrasound survey participants with the assistance of local government and health officials. Since up to 75% of inhabitants of the Tibetan plateau are illiterate, the Tibetan questionnaire was administered orally to those partaking in the survey. There were 39 individuals, identified as being echinococcosis positive via abdominal ultrasound, who consented to participate in the study. A cross-matched population (n = 39) based on age and sex, drawn from those testing ultrasound negative, was then administered the survey and the results compared with those of the ultrasound-positive subjects. Results from the Tibetan plateau echinococcosis survey were also evaluated against the standardized 1998 United States norm.6 All comparisons were made using a Student’s t-test.

Construction of DALYs. The use of DALYs is an attempt to quantify the burden of a disease, in this case echinococcosis, for Shiqu County (Sichuan Province, People’s Republic of China). The basic formula for DALYs lost by an individual is as follows

\[ \text{DALY} = \frac{DCe^{-rL}}{\beta + r} \left[ e^{(\beta+r)L} - (1 + (\beta + r)(L + a)) - (1 + (\beta + r)a) \right] \]

where, \( r \) is a discount rate, \( \beta \) is an age-weighting parameter, \( C \) is an age-weighting correction constant, \( D \) is a disability weight, \( a \) is the age of the individual at diagnosis, and \( L \) is the time lost to disability or premature mortality.7 Parameter values used were \( r = 3\% \), \( \beta = 0.04 \), and \( C = 0.16243 \).7,8 Disability weights (\( D \)), derived for AE and CE, were based on values for liver cancer obtained from the original Global Burden of Disease Study as well as from the Dutch Disability Weight Group, which produced a set of disability weights for use in a western European context.19 Liver cancer was chosen for this purpose since, like echinococcosis, it causes a space-occupying mass and often results in similar clinical symptoms (Table 1).17–20

Life expectancy was based on the Japanese estimated life span, which is one of the longest known, and was used to standardize DALYs lost in accordance with the Global Burden of Disease Study.7 A life expectancy of 82.50 years was, therefore, chosen for females and 80.0 years was chosen for males. A model life-table, West Level 26, was used to estimate expected longevity for each age, with a Chinese life-table used for comparison.7,21 The general DALY formula was used in the construction of DALYs specific for AE and CE. The DALYs were constructed on the premise of solely chemotherapeutic therapy because this is the most common treatment modality for the region and in nearly all cases the only treatment currently available.

Analysis. A DALY for AE was developed with disability outcomes divided into five components (cured, improved, stable, worse, or death) based on the health survey as well as findings from past studies in which albendazole was used as the sole treatment of human AE (Table 2).22–26 To model uncertainty, Monte Carlo techniques were used using PopTools software (Commonwealth Scientific and Industrial Research Organization, Sydney, Australia). From published data (Table 2), the results of chemotherapeutic treatment of 103 AE patients were used to construct a multinomial distribution for the likely outcome of treatment. Of these 103 subjects, there was an approximate probability of 4% of cure resulting from calcification and regression of the lesions. Patients in this category were assigned a disability weight of 0.200 (Dutch weight for clinically disease free cancer) for five years. A probability of approximately 27% was given for having mild disease (improved) with disability weight 0.200 (Dutch weight for clinically disease free cancer), a probability of approximately 41% was given for having disease equated to a disability weight of 0.239 (stable) (the Global Burden of Disease weight for pre-terminal liver cancer), and a probability of approximately 22% was given for severe disease equating to a disability weight 0.809 (worse) (the Global Burden of Disease weight for terminal liver cancer). Patients assigned to these three disease states were provided with a disability weight until the end of their expected lifespan based on a trinomial distribution. In addition, approximately 6% of the

<table>
<thead>
<tr>
<th>Table 1</th>
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<tbody>
<tr>
<td><strong>Comparison of the presenting clinical signs of alveolar echinococcosis (AE) and cystic echinococcosis (CE) of the liver with hepatocellular carcinoma (HCC)</strong></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Presenting clinical signs</th>
<th>Jaundice</th>
<th>Hepatomegaly</th>
<th>Mass-related pain</th>
<th>Lung involvement</th>
<th>Asymptomatic*</th>
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<tbody>
<tr>
<td>HCC (n = 336)</td>
<td>42.6%</td>
<td>83.9%</td>
<td>56%</td>
<td>3.2%</td>
<td>2.1%</td>
</tr>
<tr>
<td>AE (n = 30)</td>
<td>43%</td>
<td>23%</td>
<td>20%</td>
<td>3%</td>
<td>7%</td>
</tr>
<tr>
<td>AE (n = 76)</td>
<td>25%</td>
<td>14%</td>
<td>25%</td>
<td>7%</td>
<td>14%</td>
</tr>
<tr>
<td>AE (n = 33)</td>
<td>21%</td>
<td>76%</td>
<td>60%</td>
<td>9%</td>
<td>18%</td>
</tr>
<tr>
<td>CE (n = 59)</td>
<td>7%</td>
<td>5%</td>
<td>42%</td>
<td>9%</td>
<td>36%</td>
</tr>
</tbody>
</table>

* These cases were found incidentally in patients without clinical signs (diagnosed by chance at necropsy, laparotomy, or during ultrasound examination for other reasons such as pregnancy). Other categories were diagnosed clinically and confirmed radiologically.
patients were assigned the outcome of eventual death, which equates to a disability weight of 0.809 for 10 years followed by death. Using these probabilities, subjects from a population of 103 were repeatedly and randomly assigned to these five groups with the above probabilities to model the uncertainty associated with the results from a sample size of 103. Thus for AE, the proportion \( a_{AE} \) assigned to the cure category varied as \( a_{AE} \sim \text{multinomial}(103, 0.04) \), the proportion \( b_{AE} \) with disability weight 0.200 varied as \( b_{AE} \sim \text{multinomial}(103, 0.27) \), the proportion \( c_{AE} \) with disability weight 0.239 varied as \( c_{AE} \sim \text{multinomial}(103, 0.41) \), the proportion \( d_{AE} \) with disability weight 0.809 varied as \( d_{AE} \sim \text{multinomial}(103, 0.22) \), and the proportion \( e_{AE} \) assigned death in 10 years varied as \( e_{AE} \sim \text{multinomial}(103, 0.06) \), where \( a_{AE} + b_{AE} + c_{AE} + d_{AE} + e_{AE} = 1 \).

Disability weights for CE were assigned in a similar manner based on the results of albendazole treatment of 547 patients from past studies (Table 3).22-24,27-28 There were no fatalities reported in these studies due, in part, to the absence of long-term follow-up. Therefore, an approximate 1% fatality rate was assigned to account for cases that will likely progress. Therefore, the proportion \( a_{CE} \) assigned to the cure category varied as \( a_{CE} \sim \text{multinomial}(547, 0.47) \), the proportion \( b_{CE} \) with disability weight 0.200 varied as \( b_{CE} \sim \text{multinomial}(547, 0.35) \), the proportion \( c_{CE} \) with disability weight 0.239 varied as \( c_{CE} \sim \text{multinomial}(547, 0.13) \), the proportion \( d_{CE} \) with disability weight 0.809 varied as \( d_{CE} \sim \text{multinomial}(547, 0.04) \), and the proportion \( e_{CE} \) assigned death in 10 years varied as \( e_{CE} \sim \text{multinomial}(547, 0.01) \), where \( a_{CE} + b_{CE} + c_{CE} + d_{CE} + e_{CE} = 1 \).

A uniform distribution was used to subtract between 0 and 5 years from the age of abdominal ultrasound diagnosis to model the age of onset of morbidity rather than the age of detection provided by the ultrasound diagnosis. The uncertainty of the point prevalence estimates was modeled using a binomial distribution. Thus, the prevalence rate \( P_{AE} \) in the general population for AE was modeled as \( P_{AE} \sim \text{multinomial}(N_t, N_{AE}/N_t) \), where \( N_t \) is the sample size that undertook ultrasound examination and \( N_{AE} \) were the adjusted number positive for AE. The prevalence rate \( P_{ACE} \) of abdominal CE was modeled as \( P_{ACE} \sim \text{multinomial}(N_t, N_{ACE}/N_t) \), where \( N_{ACE} \) is the adjusted number that were positive for CE on abdominal ultrasound. In both cases of AE and CE, \( N_t = 3,135 \) (see above).

In addition, past studies have indicated that only approximately 75% of CE cysts are located in the liver, with a substantial proportion of cases suffering from pulmonary echinococcosis, which cannot be diagnosed with ultrasound techniques.29 Therefore, to account for pulmonary and other cases of non-diagnosed CE, the prevalence of CE for the improved/stable/worse category was modeled as: \( P_{CE} = P_{CE} \times \Gamma \), where \( \Gamma \sim \text{uniform}(1.17, 1.33) \).

A spreadsheet model incorporating the DALY formula was constructed in Excel® (Microsoft, Redmond, WA). Monte Carlo routines were implemented to re-sample across the distributions 10,000 times to account for variability in disability weight and prevalence estimates. Individuals diagnosed with both AE and CE were categorized as having AE for analysis purposes. Disability weight assignment was assumed to be age independent since there is no evidence to suggest disparity in clinical presentation dependent on age of onset. Since the prevalence of AE and CE vary at the township level, an average was taken for the entire county and the age and sex distribution of patients identified in this study was applied at the county level.

### RESULTS

The 39 questionnaires for echinococcosis-positive participants were completed in their entirety except for one missing response for each of the following questions: 3a, 3b, 4a, 4b, 5, 6a, 6b, and 6c. There were no missing responses for the control group. Of the 39 positive individuals, 26 (67%) were female and 13 (33%) were male. Patient ages ranged from 8 to 80 years. Of the females, 60% were less than 41 years of age and 39% of males were less than 41 years of age. Patients with CE made up 51% of the echinococcosis-positive group and patients with AE made up the other 49%. If an individual did not answer a question, the domain the question contributed to was not included in analysis for that person. Based on comparison of mean scores for the Shiqu County control

<table>
<thead>
<tr>
<th>Number in study</th>
<th>Cured</th>
<th>Improved</th>
<th>Stable</th>
<th>Worse</th>
<th>Death</th>
<th>Reference</th>
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<tbody>
<tr>
<td>5</td>
<td>0</td>
<td>1 (20%)</td>
<td>2 (40%)</td>
<td>1 (20%)</td>
<td>1 (20%)</td>
<td>22</td>
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<tr>
<td>11</td>
<td>2 (18%)</td>
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<td>5 (46%)</td>
<td>3 (27%)</td>
<td>1 (9%)</td>
<td>23</td>
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<tr>
<td>35</td>
<td>2 (6%)</td>
<td>4 (11%)</td>
<td>25 (72%)</td>
<td>4 (11%)</td>
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<td>24</td>
</tr>
<tr>
<td>37</td>
<td>0</td>
<td>11 (30%)</td>
<td>10 (27%)</td>
<td>12 (32%)</td>
<td>4 (11%)</td>
<td>25</td>
</tr>
<tr>
<td>15</td>
<td>1 (7%)</td>
<td>12 (80%)</td>
<td>0</td>
<td>2 (13%)</td>
<td>0</td>
<td>26</td>
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</tbody>
</table>

<table>
<thead>
<tr>
<th>Number in study</th>
<th>Cured</th>
<th>Improved</th>
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<th>Worse</th>
<th>Death</th>
<th>Reference</th>
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<tr>
<td>58</td>
<td>14 (24%)</td>
<td>29 (50%)</td>
<td>15 (26%)</td>
<td>0</td>
<td>0</td>
<td>22</td>
</tr>
<tr>
<td>253</td>
<td>72 (28%)</td>
<td>129 (51%)</td>
<td>46 (18%)</td>
<td>6 (3%)</td>
<td>0</td>
<td>24</td>
</tr>
<tr>
<td>59</td>
<td>50 (85%)</td>
<td>5 (8%)</td>
<td>1 (2%)</td>
<td>3 (5%)</td>
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<td>27</td>
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<tr>
<td>118</td>
<td>97 (82%)</td>
<td>6 (5%)</td>
<td>0</td>
<td>15 (13%)</td>
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<td>27</td>
</tr>
<tr>
<td>59</td>
<td>25 (42%)</td>
<td>25 (4%)</td>
<td>9 (16%)</td>
<td>0</td>
<td>0</td>
<td>28</td>
</tr>
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</table>
group and the United States norms, it was decided that the echinococcosis-negative group from Shiqu County was a better indicator of the standard to which echinococcosis-positive patients in the area should be compared. The CE-positive individuals were compared with AE-positive individuals for all eight domains of health. No significant difference was found ($P > 0.05$) between the two groups for any of the domains tested and it was, therefore, decided to combine the CE-positive and AE-positive persons into a single echinococcosis-positive group for further analysis.

Individuals with a positive diagnosis of *E. multilocularis* or *E. granulosus* infection had a significantly lower mean score for all eight areas of health (physical functioning, role-physical, bodily pain, general health, vitality, social functioning, role-emotional, and mental health) and the two component scores (PCS and MCS) compared with the cross-matched population from the same region ($P < 0.05$) (Figure 1). Males and females from Shiqu County were compared with an analysis for sex bias. Scores in all areas were within one standard error for both the control group and the echinococcosis-positive group. When echinococcosis-negative males were compared with echinococcosis-positive males, the control group scored higher in all categories ($P < 0.05$) except social functioning. Echinococcosis-negative females scored significantly higher than echinococcosis-positive females in all categories ($P < 0.05$). Individuals less than 41 years of age were compared with individuals greater than 40 years of age. When control groups and echinococcosis-positive groups were evaluated, it was shown that the older group, on average, scored the same or lower than the younger age group in all areas except for the vitality domain for the control group and the mental health domain for the echinococcosis-positive group. The only categories showing a significant difference, however, were bodily pain and role-emotional for the control group. Due to the findings of the SF-12 version 2 health survey, it was confirmed that human echinococcosis was associated with a decrease in the overall quality of life.

Of the 3,135 subjects examined with abdominal ultrasound, 178 cases were positive for CE (5.68%) and 180 cases (5.74%) were positive for AE (Figure 2). The distribution by age and sex of the screened population and total population is shown in Figure 3, with the total proportion of the screened population infected given in Figure 2. The estimated total adjusted prevalence was 4.6% for AE and 4.9% for CE, with an overall adjusted prevalence rate of 9.5%. Using the estimated adjusted variation of prevalence with age in Shiqu County and the West Level 26 life table, we estimated an echinococcosis burden of disease estimate of 50,933 (95% confidence interval [CI] = 41,995–61,026) DALYs lost the region (Figure 4). Alveolar echinococcosis contributed 32,978 (95% CI = 25,019–42,422) DALYs and CE contributed 17,955 (95% CI = 14,268–22,128) DALYs to the total value. This represents a loss of approximately 0.81 DALY per resident of Shiqu County due to echinococcosis or approximately 0.085 per person per 1% prevalence. When the data were fit to a Chinese life-table with a life expectancy of 68.85 years for males and 72.99 years for females, the total estimated DALYs lost for Shiqu county was 49,601 (95% CI = 40,781–59,446) (Figure 4).

**DISCUSSION**

Since disability weights have never before been assigned to human *Echinococcus* infection, it was therefore necessary to apply weights using the resources available to this study. A health survey was decided upon as one of the most attainable ways of showing a decrease in overall health of individuals with echinococcosis compared with the population norm. The SF-36 health survey has been used to indicate differences in health status between echinococcosis-positive individuals compared with a local cross-matched population. In Jordan, individuals treated for CE scored significantly lower in the role physical and bodily pain categories, which was used as a justification for including morbidity costs in an economic model for the same region. A similar study conducted in Wales showed a reduction in quality of life of individuals treated surgically for CE. In contrast to the Shiqu County study, the Jordanian and Welsh participants had been treated for and were aware of their condition and its potential outcome. The physical impact of abdominal surgery may also have contributed to these patients’ overall change in quality of life. In contrast, the Shiqu County study allows for a pre-treatment evaluation of the association of morbidity with the condition itself. The SF-12 version health survey results for this study confirm that morbidity associated with echinococcosis needs to be considered, but do not prove that echi-

![Figure 1](image1.png)  
**Figure 1.** Mean Health Scores from the short form 12 version 2 health survey for echinococcosis-positive (Pos.) patients versus a control group from Shiqu County, Sichuan Province, People’s Republic of China. Error bars show the standard error of the mean.
Echinococcosis caused the decrease in the recorded quality of life. It is possible that subjects with a low quality of life are more susceptible to infection.

DALYs were decided upon as the most suitable measure of disease burden for this study, even though there has been controversy over the appropriateness of their use in the past.\(^{31,32}\) One issue is the use of a single life table, based on the Japanese life span, being used over a vast range of populations where life expectancy may not be as high. In this study, using a Chinese life table resulted in a 2.6% decrease in the total number of DALYs lost due to echinococcosis. Another criticism directed at the DALY is that it assigns global disability rates without allowing for cultural or socioeconomic differentiation between tested populations.\(^{33}\) The DALY, therefore, most likely undervalues the true disability caused by diseases and disabilities in developing countries. Others have argued that the DALY devalues the life of a disabled person and that age-weighting also devalues the lives of individuals on an un-grounded basis.\(^{31,34}\) Even with these acknowledged obstacles, the DALY is still widely used and is generally acknowledged as one of the best ways in which to quantify an estimated measurement of morbidity and mortality from a given disease for a given population.

Deciding whether to incorporate mortality and cure into the DALY was another complexity, seeing that length of illness associated with AE and CE is extremely variable depending on location of the lesion or lesions as well as the rate at which the cyst grows or metastasizes. Without the benefit of surgical or chemotherapeutic treatment, the maximum life expectancy, after the time of diagnosis, for an AE patient is approximately 15 years.\(^ {35,36}\) In contrast, CE patients have the potential to live an extended period of time, with one case study reporting a patient who lived with latent CE for 53 years.\(^ {37}\) The mortality rate for untreated CE, however, is not

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**Figure 2.** Proportion of the screened population infected by age and sex (Shiqu County, Sichuan Province, People’s Republic of China). The upper graph represents cases of alveolar echinococcosis (with 95% exact binomial confidence limits) and the lower graph represents cases of cystic echinococcosis (with 95% exact binomial confidence limits). Females are represented in gray and males are represented in white.
known although operative fatality is estimated at approximately 2% or less.\(^1\) Long-term fatality rates associated with CE and AE treated solely with albendazole also remain unknown since chemotherapy with benzimidazoles is still a relatively recent development. In addition, spontaneous calcification of lesions and cure as well as albendazole associated calcification and cure of both CE and AE have been reported and, therefore, included in the DALY estimation.\(^{14,38,39}\)

Distribution of disability weights, consequently, proved to be challenging due to the varying clinical outcomes of the diseases, as well as the fact that methods used for assigning these weights in the past remain quite vague.\(^8\) Disability weights for AE and CE were, therefore, assigned based on preceding articles reporting success and failure of treatment exclusively with albendazole. These reports were used only as a guideline, however, since many patients in these studies were deemed unlikely surgical candidates. In addition, previously reported studies have only followed patients for a short period of time and true disease-related death rates for these patients are likely to be greatly underestimated. The closest disease state for which a DALY was constructed for the Global Burden of Disease Study was liver cancer. Values for various stages of liver cancer were taken from both the Global Burden Disease Study as well as from the Dutch Disability Weight Group and applied to AE and CE. Although echinococcosis is a more chronic disease, the similar clinical symptoms justifies using these weights. However, echinococcosis would have fewer DALYs lost if compared with a population with a similar incidence of liver cancer due to the longer life expectancy of individuals with echinococcosis.

Assigning disability weights for AE and CE was also complex due to the large number of possible outcomes both with and without treatment as well as a wide range of primary lesion sites for CE. Not all CE cases become symptomatic and spontaneous cure has been reported due to calcification of the cyst, rupture of the cyst into the bile duct or bronchial tree with subsequent expulsion of the cyst material, or via collapse and resolution of the cyst.\(^1\) In addition, CE cases with pulmonary cysts, which cannot be diagnosed via ultrasound, need to be taken into account.\(^{29,40}\) This is especially true for high altitude areas, such as the Tibetan plateau, where lung-associated disease could be more clinically severe. Therefore, even when taking into account pulmonary CE, the estimated DALYs lost remains a conservative estimate. Unlike most

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**Figure 3.** Distribution by age and sex of the screened population of Shiqu County, Sichuan Province, People’s Republic of China versus the total population. The upper graph represents females and the bottom graph represents males.
studies that have calculated the burden of other diseases, this report has attempted to take into account the uncertainty surrounding the data used to estimate disability weights and the prevalence rates of the diseases. By modeling this uncertainty using Monte-Carlo techniques, the construction of a probability density for the total number of DALYs lost has been achieved. Therefore, the assumptions described in this report are accounted for in the results given the uncertainty in the parameters. Such a stochastic approach is more useful than a deterministic approach calculating a single value for a point estimate because it gives an idea of the accuracy of the estimate of DALYs lost. The information obtained can then be used to assess the cost effectiveness of designing public health programs to control echinococcosis and to assess the risk of a poor return of DALYs saved for investment in such control programs.

The number of DALYs lost due to echinococcosis in this region is very high especially when acknowledging the potential undervaluation of DALYs in less developed parts of the world, such as the Tibetan plateau. The DALYs lost due to echinococcosis in Shiqu County is approximately 0.81 per person and compares unfavorably to the average DALY lost of 0.18 from the general Chinese population due to all disabilities evaluated combined, communicable and noncommunicable. Findings for Shiqu County are, however, not typical for China. Shiqu County and its surrounding counties are especially prone to a high prevalence of echinococcosis due to the poor socioeconomic situation, local religious beliefs and customs, and the animal husbandry practices of the region. Poor hygiene in addition to a close relationship with dogs, which have ready access to small mammals as well as offal from yaks, sheep, and goats helps contribute to the high

**FIGURE 4.** Frequency distribution of likely disability adjusted life years (DALYs) lost due to echinococcosis in Shiqu County, Sichuan Province, People’s Republic of China using a Japanese life-table (upper graph) and a Chinese life-table (lower graph).
prevalence of disease in humans. This study has clearly shown that the impact of DALYs lost due to echinococcosis, in terms of medical treatment costs, lost income, and physical and social suffering, is likely to be substantial. In addition, control options need to be considered to most efficiently decrease the incidence of AE and CE in the local population as well as decrease economic losses from *E. granulosus* infection in sheep, goats, and yaks. These issues will be addressed in a future publication.

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