A retrospective study on burden of human echinococcosis based on Hospital Discharge Records from 2001 to 2009 in Sardinia, Italy

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ARTICLE INFO

Article history:
Received 16 June 2011
Received in revised form 7 May 2012
Accepted 13 May 2012
Available online 24 May 2012

Keywords:
Cystic echinococcosis
Hospital Discharge Records
DALYs
Sardinia
Italy

ABSTRACT

Cystic Echinococcosis (CE) is an infective zoonosis that represents a worldwide important public health problem. In humans, its manifestations may range from asymptomatic infection to severe disease and possible death, and lead to economic losses from treatment costs and lost wages. Recent studies suggest that this disease has a large social impact in endemic areas, and estimates of burden in terms of monetary and non-monetary impact on human health are essential to allocate financial and technical resources. In Sardinia, the most affected Italian region per number of inhabitants, CE is still endemic, although three eradication campaigns have been carried out in 1962, 1978, and 1987, respectively. To date, the burden of human CE in Sardinia remains poorly defined. In this work, a retrospective study was carried out using public Hospital Discharge Records spanning from 2001 to 2009. During these years, a total of 1409 discharges were recorded: 1196 (84.88%) records corresponding to patients hospitalized for symptoms directly correlated to CE (primary diagnosis), and 213 (15.11%) records corresponding to patients hospitalized for symptoms not directly correlated to CE and with an afterwards or concurrent diagnosis of echinococcosis made during the hospitalization (secondary diagnosis). The annual regional average record (discharge rate) was 3.9/100,000 inhabitants. Direct cost associated with diagnosis, surgery or chemotherapy, medical care, and hospitalization in humans were evaluated in this work. Furthermore, burden of disease was also evaluated by using the disability-adjusted life years (DALYs), the preferred disease-burden measure of the World Health Organization. Knowing the burden of human CE in Sardinia is extremely important to enable the prioritization of control measures for this preventable neglected disease. This is the first study describing the measure of the overall disease burden in an Italian region endemic for this disease, performed by calculating the number of CE patients from Hospital Discharge Records.

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1. Introduction

Cystic Echinococcosis (CE) is a severe zoonosis caused by the larval stage of the cestode Echinococcus granulosus. The parasite’s life cycle is maintained through dogs, the primary definitive hosts which harbour the adult worm in their small intestine (Budke et al., 2006), and a range of domestic livestock that serve as intermediate hosts. E. granulosus eggs are excreted in the faeces of infected dogs and may contaminate soil, grass, and water. Ungulates (hoofed animals) can become infected by grazing on pastures contaminated with dog faeces. Ingested eggs hatch inside the intestine, penetrate the gut wall, and are carried by the bloodstream to different organs and tissues (mainly liver and lungs), where they develop into cysts (metacestodes) that can eventually cause severe pathological damage (Thompson and McManus, 2001). Humans can become infected by ingesting eggs that contaminate food or water, or from contact with infected dog faeces; the outcome of a human infections is the development of cysts in the liver, lungs, or other organs. Dogs usually acquire the infection from cyst-carrying livestock (especially sheep) as a result of their deliberate feeding of infected offal (liver and lungs) by owners who practice homestead slaughter, or by the occasional feeding on carcasses abandoned in the fields (Pawlowksi et al., 2001; McManus et al., 2003; Eckert and Deplazes, 2004; Benner et al., 2010). Owing to the CE cycle peculiarities, many factors contribute to persistence of the disease: high numbers of stray dogs, illegal home slaughtering, feeding dogs with infected viscera, and absence of public health education programmes (Seimens, 2003). The communities involved in sheep farming show the highest rates of infection, demonstrating
the importance of the sheep–dog cycle in transmission to humans (McManus et al., 2003).

The disease has a world–wide distribution, with endemic regions in many countries of the Mediterranean basin, Northern and Eastern Africa, Western and Central Asia, China, South America and Australia (Seimenis, 2003; Jenkins et al., 2005; Sadjadi, 2006; Yang et al., 2006; Goulugur et al., 2009). In Europe, the most affected regions are some areas of Spain, Southern Italy and Sardinia, where the annual incidence rates in humans reach 4–8/100,000 inhabitants (Eckert et al., 2002; Romig et al., 2006; Benner et al., 2010). Three echinococcosis eradication campaigns have been carried out in Sardinia, in 1962, 1978, and 1987, respectively. Over the years 1969–1990, it became clear that the Sardinian hydatid control programme had little or no effect, and that CE incidence in humans did not change significantly from the range of 13.4–22.2/100,000 inhabitants, observed in different provinces of Sardinia for the years 1948–1970 (Craig and Larrieu, 2006). To date, this parasitic disease is still endemic in the island.

Human cystic echinococcosis is a cause of substantial morbidity and mortality worldwide, and it is responsible for significant economic losses in the public health sector (Craig and Larrieu, 2006). CE may have various consequences, including direct monetary costs for diagnosis, hospitalization, surgical or percutaneous treatments, post-treatment care, and logistic, for both patient and family members, as well as indirect costs for mortality, suffering and social consequences of disability, loss of working days or "production", and abandonment of farming or agricultural activities by affected or at-risk persons (Craig and Larrieu, 2006; Battelli, 2009). The most obvious is the cost for expensive medical or surgical treatment in human cases. In addition, recent quality of life surveys suggest that patients treated for CE never fully recover and have a significant and permanent decrease in the quality of life. This has yet to be translated into monetary terms, but it will almost certainly result in income losses, lower paid jobs, and/or additional expenses due to ill health (Torgerson, 2003).

Disability-adjusted life years (DALYs) have emerged in the international health policy lexicon as a measure of the ‘burden of disease’ (total health, socioeconomic, and financial cost of a given disease to society) (Murray, 1994). The societal burden of disease, in terms of DALYs, takes into account prevalence of the disease, duration, age and gender distribution of patients, and life expectancy at the time of diagnosis. The health gap measured in terms of DALYs extends the concept of potential years of life lost due to premature death, in order to include equivalent years of “healthy” life lost in states of less than full health, broadly termed disability. One DALY is thus one lost year of healthy life. This measure is currently the preferred disease-burden measure of the World Health Organization (Carabin et al., 2005). Therefore, the main goal of this study was to estimate the burden of human CE in Sardinia. We used the Hospital Discharge Records (HDRs) drawn from a regional database provided by the Regional Department of Health, during a 9-year period from January 1, 2001 to December 31, 2009, to update the discharge and disease rates of human CE in the region. Analysis of the HDRs was the prerequisite for estimating the overall socioeconomic impact and the burden of human CE in the period under study. Such an estimate is imperative since it can be used as a tool to prioritize control measures for this largely preventable neglected disease.

2. Materials and methods

2.1. Study area

Sardinia is the second largest island in the Mediterranean Sea, with a surface of 24,089 km$^2$. It is the fourth least populated region in Italy with 1675,411 inhabitants in 2009, having a low mean population density of 69 inh./km$^2$ compared to 200 inh./km$^2$ in Italy. In the study period (2001–2009) the population growth was of 33,732 inhabitants (Istituto Italiano di Statistica, 2010). The larger urban centres are located near the coastline, while the interior is sparsely populated. Farming is still of outstanding importance, especially sheep rearing; in the island there are 43,877 farms, of which 11,356 are sheep farms with a total of 3,279,420 sheep, corresponding to half of the total Italian stock; moreover, goat farms are 1973 with 281,999 animals, cattle farms are 9496 with 264,075 animals, horse farms are 3982 with 9789 animals, and swine farms are 14,991 with 199,751 animals (National Animal Identification Database). The animal density peaks to 182/km$^2$ in the Province of Nuoro, located in East-central Sardinia, with a mean value of 149/km$^2$ compared to 23/km$^2$ in the Italian mainland.

Currently, data about farm dogs are unknown. Moreover, dogs are mostly unregistered, and the farmers keep several dogs for guarding livestock, free to roam, that do not receive an adequate parasiticide treatment. These owned dogs and many stray dogs are free to feed on dead animals that are not removed from the fields. The traditional home slaughtering of animals, combined with religious or local festivities, constitute ideal conditions for the spread of CE to dogs and humans (Scala et al., 2004; Garippa et al., 2004).

2.2. Data on humans

Human data were obtained from Hospital Discharge Records (HDRs) drawn from a regional database provided by Regional Department of Health, during a 9-year period: January 1, 2001 to December 31, 2009. The HDR contains data both on the Hospital, including Hospital code and department code, and on the patients, including patient’s hospital acceptance code, admission and discharge date, admission and discharge ward, gender, date of birth, age, domicile code, primary and secondary diagnosis codes, surgical or medical procedure codes, length stay in hospital, mortality data, Diagnostic Related Groups (DRG) code and Medical Disease Cluster (MDC) code. Anonymous anagographic data are available according to the current privacy regulations (L.675/96) (Gazzetta Ufficiale, 1997). A total of 1409 HDRs with diagnosis codes 122.0–122.9, based on International Classification of Diseases, 9th revision, Clinical Modification (ICD-9-CM), were processed (Center for Disease Control and Prevention, 2010). We considered records of patients hospitalized for symptoms directly correlated to CE (primary diagnosis), and records of patients hospitalized for symptoms not directly correlated to CE and with an afterwards or concurrent diagnosis of echinococcosis made during the hospitalization (secondary diagnosis). The HDRs were analysed according to the patient’s domicile code in order to evaluate the discharge rate distributed in the territory. The total number of discharge records during 2001–2009 and per year were counted. These data were calculated in Sardinia and in the provinces of Nuoro, Sassari, Cagliari and Oristano. The number of discharges per year was grouped considering ordinary hospitalization (OH), day hospitalization (DH) and surgical or medical procedure. DH refers to procedures economically supported by the public welfare but that require a stay in hospital without overnight stay. Direct costs were calculated on DRG; DRG for each record is calculated on patient’s age, gender, primary and possibly secondary diagnosis, list of treatments/procedures and average length of hospital stay. To cost out the DRG and evaluate the total direct cost, itemized prices defined by the Sardinian Regional Government (Italian Ministry of Health, 1994; Regione Autonoma della Sardegna, 2011) were used.
2.3. Disability-adjusted life years (DALYs)

When estimating the health gap for the years lived by an individual in a state of ill health, disabilities are modulated by non-uniform age weights in order to reflect social preference for years of healthy life lived by young adults. The weight \( e^{-\beta t} \) (where \( t \) is the time and \( \beta > 0 \) is a constant) attains its maximum for \( t = 1/\beta \). Usually, \( \beta = 0.04 \) is chosen, corresponding to a maximum at age 25 (Budke et al., 2006). Moreover, following the standard economic principle of discounting future benefits, current discount rates are applied to disability in the future. Here we used the discount rate \( r = 3\% \). For \( L \) years lived with disability \( D \) by one individual since age \( a \), DALY (\( r, 1 \)) (that is, with discount rate \( r \) and non-uniform age weights) equals

\[
C \int_0^{+\infty} D t e^{-r(t-a)} dt = \frac{CD e^{-\beta a}}{(\beta + r)} \left[ e^{-(\beta + r)(L + a)} - (1 + (\beta + r)a) \right]
\]

with a normalization constant \( C = 0.1658 \) such that DALYs with uniform and non-uniform age weights are the same (Benner et al., 2010; Craig and Larrieu, 2006). More precisely, this formula gives YLL (Years of Life Lost) for fatal outcomes (where \( D = 1 \) and \( L \) is the estimated remaining lifespan) and YLD (Years Lived with Disability) for nonfatal outcomes. Then DALY = YLL + YLD and, finally, DALYs for a given sequela of a given illness are the sum of individual DALYs. Duration \( L \) and disability coefficient \( D \) for echinococcosis are not deduced from hospital records but are instead chosen according to the Budke and Stouthard rule (Seimenis, 2003; Stouthard et al., 2000), and applied to groups of patients. Disability is divided into classes as defined by Murray (1994), with each class having a severity weight between 0 and 1.

3. Results

Between 2001 and 2009, 1409 HDRs with CE related diagnoses codes 122.0–122.9 were collected from all Sardinian hospitals and analysed. In this period, the number of records ranged from a minimum of 112 in 2005, to a maximum of 199 in 2008, with an average number of 156.5 records per year (Table 1). Of these, 1196 (84.88%) were records with a primary diagnosis, and 213 (15.11%) were records with a secondary diagnosis.

A mean annual regional discharge rate of 9.3/100,000 inhabitants emerged (Table 1). An analysis of single municipalities was then carried out, revealing that the highest discharge rate was present in the Province of Nuoro, with an annual discharge rate of 25.8/100,000 inhabitants (Table 2).

The 1409 HDRs were divided into ordinary hospitalizations (OH) and Day hospitalizations (DH) stratified per year. A total of 1266 records, corresponding to 89.8% of 1409 HDRs, were OH, against only 143 records (10.1%) of DH (Tables 3–4).

For each hospitalization event, our data report which type of treatment was applied, whether surgical (type of surgical intervention undergone) or medical (management of pharmacological/drug therapy). Most hospitalizations, 751 (53.3%) of OH and 143 (100%) of DH, were solved by means of medical care, although surgical procedures remain still noticeable in the OH group (515 or 36.5%). It is to be noted that the annual percentage of total medical treatments reached 68% in 2007, starting from 50% in 2001 (Table 3).

The data obtained upon analysis of the 1409 HDRs were used to assess the direct cost for echinococcosis in Sardinia from 2001 to 2009, using the itemized prices defined by The Sardinian Regional Government (Regione Autonoma della Sardegna, 2011) to cost out the DRG. During the reporting period, the direct cost for 1266 OH was €6,625,453 distributed as €4,561,244 (range €381,555 in 2005 and €783,628 in 2001) for 515 OH with surgical procedures and €2,064,209 (range €87,660 in 2001 and €320,444 in 2009) for 751 OH with medical care. The mean cost of a single OH was €5,316 (range €617 in 2001 and €7161 in 2002) considering an average length of hospital stay of 15.1 days (Table 3). The direct cost for 143 DH was €91,396 (range €175 in 2003 and €15,996 in 2006) with a mean cost for each of €653 (range €145 in 2003 and €143 in 2009) (Table 4). From 2001 to 2009 the total direct cost (OH plus DH) for echinococcosis in Sardinia was €6,716,849 corresponding to a mean cost of €76,416 per year. Mean direct costs of treating a case of human echinococcosis in Sardinia have been calculated to be €5970.

To compute the burden of disease in terms of DALYs, we calculated how many patients, identified in records by domicile code analyses, gender and age at the time of diagnosis, were hospitalized at least once in the whole period: the total of 1409 records corresponded to 938 single patients, of which 667 (71%) patients hospitalized once and 271 (29%) patients with multiple hospitalizations. Patients hospitalized once were included into 71.10% of patients improving after surgery or medical care, accounting to 313 and 354, respectively; 271 patients with multiple stays were grouped in 225 (23.98%) patients with less than 3 and more than 1 stay (patients developing morbidity) and 46 (4.9%) patients with more than 3 stays, of which 22/46 (2.3%) patients with medical treatment (developing relapse after medical care) and 24/46 (2.5%) for surgical procedure (developing relapse after surgery) (Table 5) (Murray, 1994).

For each patient group, the average age was calculated and different parameters of disability (D) and duration (L) were applied with the above percentages of patients (Table 5) (Murray, 1994). The evaluation of the burden of disease, in terms of DALYs, takes into account the disease prevalence, the duration in terms of number of hospitalizations and relapses, medical treatments and surgical procedures, age distribution and the remaining life expectancy at the age of onset.

Total DALYs were 505.40 with a range of 169.32 in the age group 41–60 and 20.76 in the age group 81+90. For each age group and patient group, the DALY value was calculated separately (Table 6). The group of 225 patients developing morbidity and with less than 3 and more than 1 hospitalization, showed the higher DALY value. The age group with the higher DALY value was 41–60.
Table 3
Number of Diagnosis Related Groups (DRGs) of ordinary hospitalization per year, grouped by surgical procedure (S), medical care (M), average day of hospitalization and direct cost in euro (€). Direct costs are averages per single OH and totals per year during 2001–2009.

<table>
<thead>
<tr>
<th>Year</th>
<th>OH-DRG</th>
<th>Average days of hospitalization</th>
<th>Mean direct cost (€) per single OH</th>
<th>Direct cost (€) per year</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>S</td>
<td></td>
<td></td>
<td>S</td>
</tr>
<tr>
<td>2001</td>
<td>94</td>
<td>17.0</td>
<td>4978.78</td>
<td>783,628.43</td>
</tr>
<tr>
<td>2002</td>
<td>69</td>
<td>19.5</td>
<td>7161.37</td>
<td>760,694.87</td>
</tr>
<tr>
<td>2003</td>
<td>54</td>
<td>16.0</td>
<td>6369.69</td>
<td>533,312.84</td>
</tr>
<tr>
<td>2004</td>
<td>65</td>
<td>17.5</td>
<td>5883.71</td>
<td>588,609.18</td>
</tr>
<tr>
<td>2005</td>
<td>43</td>
<td>16.0</td>
<td>6052.48</td>
<td>381,555.77</td>
</tr>
<tr>
<td>2006</td>
<td>50</td>
<td>11.5</td>
<td>4171.43</td>
<td>415,774.25</td>
</tr>
<tr>
<td>2007</td>
<td>52</td>
<td>12.0</td>
<td>4555.64</td>
<td>447,561.51</td>
</tr>
<tr>
<td>2008</td>
<td>59</td>
<td>12.0</td>
<td>4244.78</td>
<td>473,965.75</td>
</tr>
<tr>
<td>2009</td>
<td>28</td>
<td>14.0</td>
<td>4433.78</td>
<td>174,142.08</td>
</tr>
<tr>
<td>2001–2009</td>
<td>515</td>
<td>Mean 15.1</td>
<td>Mean/yr 5316.85</td>
<td>64,561,244.00</td>
</tr>
<tr>
<td></td>
<td>1266/1409</td>
<td></td>
<td></td>
<td>€2,064,209.40</td>
</tr>
</tbody>
</table>

Table 4
Number of diagnosis related groups (DRGs) of Day Hospitalization per year, grouped by medical care (M) and direct cost in euro (€). Direct costs during 2001–2009 are reported as averages per single DH and totals per year.

<table>
<thead>
<tr>
<th>Year</th>
<th>DH DRG</th>
<th>Mean direct cost (€) per single DH</th>
<th>Direct cost (€) per year</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>S</td>
<td></td>
<td></td>
</tr>
<tr>
<td>2001</td>
<td>12</td>
<td>479.90</td>
<td>5759.62</td>
</tr>
<tr>
<td>2002</td>
<td>19</td>
<td>446.49</td>
<td>8036.96</td>
</tr>
<tr>
<td>2003</td>
<td>15</td>
<td>345.02</td>
<td>5175.35</td>
</tr>
<tr>
<td>2004</td>
<td>19</td>
<td>538.88</td>
<td>10,234.95</td>
</tr>
<tr>
<td>2005</td>
<td>15</td>
<td>590.00</td>
<td>8850.83</td>
</tr>
<tr>
<td>2006</td>
<td>17</td>
<td>940.99</td>
<td>15,996.88</td>
</tr>
<tr>
<td>2007</td>
<td>12</td>
<td>689.54</td>
<td>8274.55</td>
</tr>
<tr>
<td>2008</td>
<td>21</td>
<td>710.37</td>
<td>14,207.5</td>
</tr>
<tr>
<td>2009</td>
<td>13</td>
<td>1143.0</td>
<td>14,859.89</td>
</tr>
<tr>
<td>2001–2009</td>
<td>143/1409</td>
<td>(Mean/yr) 653.77</td>
<td>€913,396.53</td>
</tr>
</tbody>
</table>

Table 5
Percentages of patient group (%) classified according to disability (D) and duration of disability (L).

<table>
<thead>
<tr>
<th>Patient group</th>
<th>Point estimate (%)</th>
<th>95% Confidence interval</th>
<th>Disability (D)</th>
<th>Duration (L)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Improving after surgery or medical care</td>
<td>71.1</td>
<td>68.0–74.0</td>
<td>0.200</td>
<td>1 year</td>
</tr>
<tr>
<td>Developing morbidity</td>
<td>24.0</td>
<td>21.4–26.8</td>
<td>0.239</td>
<td>5 years</td>
</tr>
<tr>
<td>Developing relapse after surgery</td>
<td>2.3</td>
<td>1.6–3.5</td>
<td>0.809</td>
<td>5 years</td>
</tr>
<tr>
<td>Developing relapse after medical care</td>
<td>2.6</td>
<td>1.7–3.8</td>
<td>0.809</td>
<td>5 years</td>
</tr>
</tbody>
</table>

4. Discussion

The Hospital Discharge Record was introduced in Italy in 1991, and it is the most important source of information for the annual public and private hospital inpatient discharge data, collected by the National Ministry of Health from all Regional Health Departments. Data collected by HDRs are relative to the Diagnosis Related Groups (DRGs) and the International Classification of Diseases, 9th revision, Clinical Modification (ICD-9-CM), which provide a means of relating the type of patients and hospital treatments to the costs incurred by the hospital (Center for Disease Control and Prevention, 2010). The analysis of HDRs by the National Ministry of Health is necessary for planning and monitoring activities and for compensation of hospital expenses.

Record of all CE patients were identified based on the procedures performed, divided into medical and surgical groups and relative to Ordinary Hospitalization (OH) and Day Hospitalization (DH). It is important to note that the data used here are related only to hospitalized patients, and do not include those attended only by private physicians and family doctors. This must be taken into account when evaluating the real economic and social impact of the disease. However, it does point out that, probably, only a minority of CE patients incur healthcare costs. This is consistent with population based data on the natural history of CE infection, that occurs

Table 6
Total DALYs by age group and outcome in the years 2001–2009 (95% confidence intervals).

<table>
<thead>
<tr>
<th>Age group</th>
<th>Improving patients</th>
<th>Patients developing morbidity</th>
<th>Patients developing relapse after surgery</th>
<th>Patients developing relapse after medical care</th>
<th>Total DALYs</th>
</tr>
</thead>
<tbody>
<tr>
<td>0–20</td>
<td>5.21–5.67</td>
<td>10.07–12.61</td>
<td>2.55–5.02</td>
<td>2.71–5.07</td>
<td>20.54–28.37</td>
</tr>
<tr>
<td>21–40</td>
<td>29.20–31.78</td>
<td>50.90–63.74</td>
<td>12.88–25.36</td>
<td>13.69–25.60</td>
<td>106.67–146.49</td>
</tr>
<tr>
<td>41–60</td>
<td>39.59–43.08</td>
<td>67.43–84.44</td>
<td>17.06–33.59</td>
<td>18.13–33.91</td>
<td>142.21–195.03</td>
</tr>
<tr>
<td>61–80</td>
<td>38.56–41.96</td>
<td>65.01–81.41</td>
<td>16.45–32.39</td>
<td>17.48–32.70</td>
<td>137.50–188.46</td>
</tr>
<tr>
<td>81+</td>
<td>4.91–5.34</td>
<td>8.24–10.32</td>
<td>2.09–4.11</td>
<td>2.22–4.14</td>
<td>17.45–23.91</td>
</tr>
<tr>
<td>Total DALYs</td>
<td>117.46–127.83</td>
<td>201.65–252.53</td>
<td>51.03–100.47</td>
<td>54.22–101.43</td>
<td>424.3–582.256</td>
</tr>
</tbody>
</table>
symptomatically in a small number of people after many years of infection and complicates estimation of the exact incidence rate. Once that the disease is established, CE requires intensive health care resources and generates very high cost, particularly in surgical patients.

HDRs from National Hospitals, obtained from the National Ministry of Health in the period 1999–2005, are a total of 9073 HDRs with an average of 1296 per year and a mean annual national discharge rate of 2.27/100,000 inhabitants. HDRs from Sardinian Hospitals are in average 156 per year, ranging from 112 records in 2005 to 199 in 2008, with a mean annual regional discharge rate of 9.3/100,000 inhabitants calculated in the last nine years. According to these data, human CE continues to be a substantial cause of morbidity in Italy and especially in Sardinia, where we calculated that 12% of the total national hospitalizations for CE occurred.

Most of the Sardinian cases were from the Province of Nuoro, with an annual discharge rate of 25.8/100,000 inhabitants (Table 2); this territory has an area of 3933.32 km² with 161,020 inhabitants and a very low population density of 37.0 inh./km² compared to 68.9 inh./km² in Sardinia and 200 inh./km² in Italy. Nevertheless, this province presents the highest density of domesticated animals, and a high percentage of inhabitants are involved in sheep farming.

The total cost of the disease is the sum of the costs of the health services, cost of morbidity and also loss in animal productivity. Human-associated economic losses that arise from diagnostic procedures, surgery and drug treatment, hospitalization, period of recovery, life impairment, and fatalities are relatively easy to identify and enable the calculation of the direct costs of patients suffering from clinical echinococcosis. The mean direct cost for treating a case of human echinococcosis in Sardinia was €5970 and the total mean direct cost per year was €746,316. Considering a national average of 1296 HDRs per year with a total of 9072 HDRs during the seven years period under examination, we can extrapolate the total direct cost in Italy being €54,174,157 with a mean annual direct cost of €7,737,120.

There are a number of other potential effects on human health that are not so readily defined. This takes the form of human morbidity and mortality costs. The question of the long term health of an individual who has either been treated for echinococcosis or is unwittingly affected by an undiagnosed disease should be considered (Torgerson, 2003). The disability-adjusted life years (DALYs) quantify the burden of disease and extend the concept of potential years of life lost to premature death, to include equivalent years of healthy life lost by virtue of being in states other than good health. When calculating indirect costs, we included also human productivity loss. In our work, patients were stratified according to gender and age groups: since the age values range from 2 to 100, we identified five groups of 20 years each. In all the patient categories under study we observed the highest value of DALYs in the age group 41–60 (169.32), where we found the second highest number of hospitalizations (28.1%). Males were more affected than females. People in this age group are probably the most active in livestock rearing, and limited ability to perform their activities has more serious consequences and is responsible for the most significant economic losses.

5. Conclusions

Notification of human CE in Italy is compulsory but widely disregarded. Furthermore, there is no national register of CE cases, and this bound us to restrict our study to hospitalized patients by using HDRs. By using the binomial exact method, it is not possible to define a precise estimate of the total number of cases, but only an estimate on uncertainty or probability of hospital discharges can be provided. The final computation of DALYs reflects this uncertainty. More accurate data on CE prevalence in humans (particularly undiagnosed or asymptomatic cases) are needed, and the activation of correct reporting measures for this infectious disease, together with the implementation of the Community Network under Decisions no. 1219/98/EC of the European Parliament and Council, is of considerable importance. This estimate complements the previous estimates on global burden of CE in other countries (Torgerson et al., 2000, 2001; Torgerson and Dowling, 2001; Majrowski et al., 2005; Benner et al., 2010).

Several years ago the WHO recommended that an economic evaluation of the effects of parasitic zoonoses should be an integral part of any control programme (World Health Organization, 1979). In Sardinia, where control programmes have been reduced due to economic problems and lack of resources, CE persists as a public health problem with a cost that is 746,316 euro per year. These data confirm the high prevalence of human echinococcosis in Sardinia. We stress that CE is a neglected and preventable zoonosis that remains a human health concern; additional funding is needed to reduce human and animal infection rates through improved disease surveillance, regular treatment of dogs and greater cooperation among agencies.

Conflict of interest

None.

Acknowledgment

We thank the Sardinia Department of Health for providing the Regional Hospital Discharge Records (HDR).

References


