Original Article

Estimation of Burden of Cystic Echinococcosis in Iran Using Disability Adjusted Life Years (DALYs) in 2018

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Abstract

Background: Human hydatidosis as a public concern has increased in a number of countries that have reduced control programs for the disease due to lack of resources or policies. We aimed to estimate Disability-Adjusted Life Years (DALYs) for human hydatidosis in Iran in 2018.

Methods: Data were collected from the Center of Communicable Diseases Control, Ministry of Health &Medical Education, Tehran, Iran in 2018. To calculate DALYs, years of life lost due to premature death (YLL) with years of life with disability (YLD) were calculated according to the formula as DALY = YLL + YLD. The standard life expectancy lost method (SEYLL) was used to calculate the years lost due to premature death.

Results: DALYs for human hydatidosis was calculated as 1210.12 years (YLD equals to 177.12 and YLL equals to 1033) in Iran for the year 2018. It was estimated to be 700.2 years for men and 509.8 years for women. DALYs in men were significantly different from women ($P= 0.001$) so DALYs were more in men than women were. YLD was calculated at 78.228 years in men and 98.892 years in women and in both men and women at 177.12 years. YLD was significantly different in women compared to men ($P=0.001$), so YLD in women was more than in men.

Conclusion: We reached considerable indices for hydatidosis in our study. Therefore, disease prevention and control programs in Iran seem necessary by the policy makers.

Keywords: Hydatidosis; Burden; Disability-adjusted life years; Human; Iran

Introduction

Cystic echinococcosis (CE) is a severe zoonotic disease that affects people all over the world. CE was included in the official list of the 18 Neglected Tropical Diseases by the WHO. In addition to

this category, the WHO has designated CE as part of a subgroup of seven endemic or "neglected zoonotic diseases" (NZDs) prevalent in areas where poverty, livestock dependency, or other factors contribute to disease transmission (1-3). Risk factors include contact with dog, eating vegetables, geophagy and contact with sheep. Ingesting embryonated eggs through hands, food, drinks or material contaminated with parasite eggs infects humans. The larvae reach the blood and lymphatic circulation and transport to the liver, lungs and other organs (2, 4). The asymptomatic period is too long and the disease might be even diagnosed after 20-25 years post infection (4).

The threat and emergence of CE in humans is a compelling reason to advance research into intervention programs to control human infections (1, 2). Iran is considered as an endemic region for hydatidosis. In addition, the trend of hydatidosis shows that it is an important health threatening infection in Iran (5, 6), so it seems necessary and urgent to develop an urgent program to monitor the diseases (4). In order to implement a sustainable hydatidosis control program, studies are needed to assess transmission and risk factors at the local level (7).

Due to demographic and geographical differences between countries, various aspects of CE, including epidemiological and clinical status, should be further investigated in each region (8). Unfortunately, recent evidence suggests that hydatidosis has become a public health problem in a number of countries where control programs have been reduced due to lack of resources or policies (9-12).

Accurate information about the disease can help the country's health decision makers in allocating resources and fighting the disease. Disability-Adjusted Life Years (DALYs) has been used by the WHO since 1990 as a more general way to quantify the burden of disease on the global burden of disease (GBD) (13). DALY is a combination of the potential life years lost due to premature death (YLL) and the years spent living with a disability (YLD) (14).

One of the benefits of using DALY as an immediate index of community health is that it allows health decision makers in each country to find out by comparing different illnesses which problem are more important or whether the condition of a disease is better or worse. For example, a study in Iran showed the mortality and high burden of traffic accidents and forced the authorities to intervene (15). Another benefit of DALY is helping to assess the state of the health system and understanding which system information sources are weak, omitted or unreliable (16). A distinctive feature of the studies performed with this method is that they provide the possibility of comparison between different diseases and regions (17).

In Iran, a few studies have been conducted on the burden of CE. Fasihi et al reported the overall annual cost of CE in Iran at US$ 232.3 million, including both direct and indirect costs. It was US$ 93.39 million for human CE and the annual cost associated with CE in livestock was estimated at US$ 132 million (18).

Therefore, considering that little attention has been paid to the burden studies of hydatidosis in Iran, this study was conducted by measuring the burden of human hydatidosis in Iran in terms of DALYs in 2018.

Methods

Data were obtained from the Center of Disease Control (CDC), Iranian Ministry of Health (MOH), Tehran, in 2018.

DALYs

The DALY index was used to calculate the load of hydatidosis. Other parameters such as disease incidence, population structure, disease mortality, and disease recovery are used to calculate disease burden (19, 20).

To calculate DALY, we first need to add the number of YLL to YLD. In order to study the epidemiological aspects of the disease in Iran, the opinions of experts in this disease have been

used. In addition, the reference of the WHO was considered (18).

YLL

The Standard Expected Years of Life Lost (SEYLL) method was used to calculate the YLL. This method uses the standard life expectancy to estimate the YLD at that age (21). Information about the deaths was obtained from the Iranian MOH Death Registration Unit, which is the gold standard for obtaining the cause of death.

To calculate the remaining potential life years at each age of death, the 2010 GBD standard life table with 86 years of life expectancy at birth was used for both sexes (22).

YLD

YLD was calculated based on the number of new cases of the disease during the disability period and the severity of the disease disability. Data on the number of new cases of the disease were obtained from the Iranian MOH.

The disease period was determined by considering the average of comments and comparison with published articles as well as the article of the WHO (23). In the 2010 GBD study, the discount rate and age weight were omitted. The weight of disability (DW) in this study was considered as 0.123 for hydatidosis according to the WHO (12, 20, 24).

Ethics Approval

Ethics Committee of Tehran University of Medical Sciences, Tehran, Iran approved the study under the code of IR.TUMS.SPH.REC.1399.030.

Statistical analysis

SPSS ver. 21 (Chicago, Il, USA) was used to analyze the data using descriptive statistics, Chi-square test and t-test. *t*-test was used to find a significant difference between the YLD, YLL and DALYs in men and women. Chi-square test was used to find a significant difference between gender and age groups concerning those indexes.

Results

The burden for human hydatidosis was calculated at 1212.12 years (YLD equals to 177.12 years and YLL equals to 1033 years), estimated to be 700.228 years for men and 509.892 years for women. According to the *t* test, the YLL in men was significantly different from women ($P=0.001$) so that DALY was more in men than women. There was a significant relationship between gender and age group in disability and death using Chi-square test ($P=0.001$). The lowest amount of DALY in men was observed in the age group of 60-69 years and the highest amount in the age group of 45-59 years. The lowest amount of DALY in women was observed in the age group of 5-14 years and the highest value in the age group of 30-44 years. Hence, the DALY estimate of hydatidosis was more affected by mortality.

DALY or and YLL were higher in men than women. YLL was calculated at 622 years in men and 411 years in women and a total of 1033 years in women and men. YLL was higher in men than women. However, statistically and using the *t* test, this difference was not significant. As for YLL using Chi-square test, a significant relationship was seen between gender and age group ($P=0.01$). In both men and women, the lowest amount of YLL was observed in the age group of 69-60 years and 5-14 years, respectively.

YLD or in other words, the burden of disability was calculated at 78.228 years in men and 98.892 years in women and in both men and women it was 177.12 years. According to the *t* test, YLD was significantly different in women compared to men ($P=0.001$) so that the burden of disability in women was more than in men. There was no significant relationship between gender and age group in YLD using chi^2 test. In both men and women, the lowest amount of YLD was observed in the age group of 0-4 years because the lowest incidence was in this age group.

Table 1 shows the epidemiological data of hydatidosis data in Iran in 2018. The number of new cases of hydatidosis was 720 cases (402 (55.83%)

for females, and 317 (44.2%) for males). The overall incidence of hydatidosis was calculated at 0.9 per 100,000 people (0.8 men and 0.99 women). The disease period was 2 years for men and women.

Table 2 shows the YLL, YLD, DALYs and DALY/1000 related to hydatidosis in Iran in 2018.

Table 1: Epidemiological data of human hydatidosis in Iran in 2018

Age group (yr)	Number of population per thousand		Number of cases		Incidence rate in one hundred thousand	
	M	F	M	F	M	F
1-5	3751	3566	1	4	0.03	0.1
5-10	3500	3321	18	19	0.5	0.6
10-15	3041	2899	23	14	0.7	0.5
15-20	2777	2656	23	19	0.8	0.7
20-25	2972	2883	31	19	1	0.6
25-30	3798	3717	30	35	0.8	0.9
30-35	4379	4302	28	36	0.6	0.8
35-40	3926	3833	29	41	0.7	1
40-45	3062	2952	29	32	0.9	1
45-50	2570	2482	13	36	0.5	1.4
50-55	2138	2101	18	33	0.8	1.5
55-60	1756	1767	21	33	1.2	1.9
60-65	1389	1428	9	25	0.6	1.7
65-70	924	1015	13	25	1.4	2.5
70-75	595	670	18	12	3	1.8
75-80	415	433	3	9	0.7	2.1
80+	545	520	11	10	2	1.9
Total	41538	40546	318	402	0.8	0.99

M=Male; F=Female

Table 2: YLL, YLD, DALYs and DALY/1000 related to hydatidosis in Iran in 2018

Age group (yr)	YLDs			YLLs			DALYs		
	M	F	M & F	M	F	M & F	M	F	M & F
0-4	0.246	0.984	1.23	84	84	168	84.246	84.984	169.23
5-14	10.086	8.118	18.204	79		79	89.086	8.118	97.204
15-29	20.664	17.958	38.622	64		64	84.664	17.958	102.622
30-44	21.156	26.814	47.97	98	137	235	119.156	145.268	282.97
45-59	12.792	25.092	37.884	148	69	217	160.792	94.92	254.884
60-69	5.412	12.3	17.712	25	21	46	30.412	33.3	63.712
70-79	5.166	5.166	10.332	69	63	132	74.166	68.166	142.332
>80	2.706	2.46	5.166	56	37	93	58.706	39.46	98.166
Total	78.228	98.892	177.12	622	411	1033	700.228	509.892	1210.12
		P= 0.001			*P = 0.101*			*P =0.001*	

M=Male; F=Female

Discussion

Despite the establishment of control programs in a number of countries or indigenous regions, hy-

datidosis is still prevalent in a wide geographical area. This disease is considered an important issue of public and economic health in Iran (4). However, information about the burden of the

disease and its economic consequences on society is not fully estimated in many endemic countries.

In western China, the burden of hydatidosis was 20,330 (the YLD was estimated to be 1812 years and, the YLL was estimated at 18,518 years (25). In Tibetan, DALYs for hydatid cysts were estimated at 17,955 which had a significant impact on the medical treatment costs, income loss, physical and social suffering (26). In Kyrgyzstan, DALYs for hydatid cyst were reported as 3,052 (27). In a review article, DALYs were reported for this disease at 183,537 (28). Again, in the Tibetan Plateau, the echinococcosis burden was reported using DALYs as 50,933 (26). The disease burden of these studies was higher than the results of our study. The difference might be mentioned as the higher rate of hydatidosis in above mentioned regions.

In Italy, from 2001 to 2014, the authors estimated DALYs for hydatid cyst at 3127.71, YLL at 1003.62, and YLD at 2124.09 (12). In Nepal, from 2000 to 2012, the burden of common parasitic diseases was calculated. Accordingly, DALYs for the most common diseases was calculated as 14268 for neurocysticercosis, 9255 for congenital toxoplasmosis and 251 for hydatid cyst (29). In 2017, DALYs was reported at 322.398 for China (30). The disease burden of these studies was lower than the result of the present study. The difference between the burden of this study and other studies is probably due to the number of cases per year and the number of deaths due to the disease in that year.

Other factors, such as race, may have an effect on the outcome of the disease, as evidenced in tuberculosis and malaria (19, 31). Significant differences in treatment according to the study area were evident in alveolar cysts or in brucellosis with inadequate treatment, most likely chronic or recurrent, which increases the load of the disease (32). Over time, improving diagnoses and using new treatments may change the outcome of the disease and eventually change the DALYs in each case. In the Kyrgyz study, for example, all cases of alveolar cysts were considered fatal due to insufficient treatment, but the Swiss study showed that adequate treatment of the disease led to increased survival (16, 27). The symptoms of the disease may not be severe and the patient may not go to health centers, or the patient with hydatid cyst may have limited access to health centers or the exact diagnosis of the disease may not be given, thus the chances of registering cases of hydatid cyst are low (27). It is worth noting that in Iran, most surgeons tend to treat human hydatidosis with chemotherapy instead of surgery or use a "watch and wait" method, and only some complex cases are operated on (discussed in internal congresses). This reduces the number of patients through hospital data (5).

Hydatid cysts are often reported to a less extent. In Uzbekistan, the number of official reported cases was 1,435 in 2000, followed by 819 cases in 2001 (33). In Uzbekistan, a total of 4,430 cases were reported in 2000, followed by 4,089 cases in 2001(33). In Chilean official records between 2001 and 2009, the average was 311 cases per year, while hospital records showed an average of 1,009 cases per year (34), indicating that discrepancies in reported data do not always indicate epidemiological changes.

One of the problems in calculating DALYs for hydatidosis is the calculation of epidemiological features, including the course of the disease (35). One of the strengths of this study was the use of the opinions of experts in this disease and all studies published in Iran. Prioritizing health care needs is inevitable for policymakers (34). DALY provides this as a tool, but alone it is not enough. Using DALY by health care policymakers, it is important to pay attention to what is being offered (36). It is also important to understand that DALY may not have the full effect of the disease and that the effects of a disease on individuals may be much more severe than those that directly affect individuals (37). Life expectancy in Iran has increased dramatically and, this trend is likely to continue (14). Therefore, it is suggested that Iranian health policymakers consider recalculating the burden of disease in the short term.

Conclusion

One of the measures taken to monitor the diseases by the policy makers is calculation of DALYs. Through our study we found the hydatidosis in Iran should be considered an important disease. Therefore, disease prevention and control programs seem necessary. We recommend recalculating this index for this disease to reach the best feature of its different aspects.

Ethical considerations

Ethical issues (Including plagiarism, informed consent, misconduct, data fabrication and/or falsification, double publication and/or submission, redundancy, etc.) have been completely observed by the authors.

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Conflict of interest

The authors declare that there is no conflict of interest.

References

1. Craig P, Hegglin D, Lightowlers M, Torgerson PR, Wang Q (2017). Echinococcosis: control and prevention. *Adv Parasitol,* 96:55-158.
2. Deplazes P, Rinaldi L, Rojas CA, et al (2017). Global distribution of alveolar and cystic echinococcosis. *Adv Parasitol,* 95:315-493.
3. Campos-Bueno A, López-Abente G, Andrés-Cercadillo AM (2000). Risk factors for Echinococcus granulosus infection: a case-control study. *Am J Trop Med Hyg,* 62:329-334.
4. Rokni M (2009). Echinococcosis/hydatidosis in Iran. *Iran J Parasitol,* 1;4(2):1-16.
5. Khademvatan S, Majidiani H, Foroutan M, Tappeh KH, Aryamand S, Khalkhali H (2019). *Echinococcus granulosus* genotypes in Iran: a systematic review. *J Helminthol,* 93:131-138.
6. Khalkhali H, Foroutan M, Khademvatan S, Majidiani H, Aryamand S, Khezri P, Aminpour A (2018). Prevalence of cystic echinococcosis in Iran: a systematic review and meta-analysis. *J Helminthol,* 92:260-268.
7. Zhou XN, Lv S, Yang GJ, Kristensen TK, Bergquist NR, Utzinger J, Malone JB (2009). Spatial epidemiology in zoonotic parasitic diseases: insights gained at the 1 st International Symposium on Geospatial Health in Lijiang, China, 2007. *Parasit Vectors,* 2:1-16.
8. Ebrahimipour M, Rezaeian S, Shirzadi MR, Barati M (2019). Prevalence and risk factors associated with human cystic echinococcosis in Iran. *J Parasit Dis,* 43:385-392.
9. Eckert J, Gemmell M, Meslin F, Pawlowski J (2001). Echinococcosis in humans and animals: a public health problem of global concern. *Manual of the World Health Organization, Geneva and of The Office International des Epizooties, Paris.*
10. Torgerson P (2003). Economic effects of echinococcosis. *Acta Trop,* 85:113-118.
11. Budke CM, Deplazes P, Torgerson PR (2006). Global socioeconomic impact of cystic echinococcosis. *Emerg Infect Dis,* 12:296.
12. Piseddu T, Brundu D, Stegel G, Loi F, Rolesu S, Masu G, Ledda S, Masala G (2017). The disease burden of human cystic echinococcosis based on HDRs from 2001 to 2014 in Italy. *PLoS Negl Trop Dis,* 11:e0005771.
13. Murray CJ, Lopez AD, Organization WH (1996). *The global burden of disease: a comprehensive assessment of mortality and disability from diseases, injuries, and risk factors in 1990 and projected to 2020: summary.* ed. World Health Organization.
14. Murray CJ, Vos T, Lozano R, et al (2012). Disability-adjusted life years (DALYs) for 291 diseases and injuries in 21 regions, 1990–2010: a systematic analysis for the Global Burden of Disease Study 2010. *Lancet,* 380:2197-2223.

15. Naghavi M, Abolhassani F, Pourmalek F, et al (2009). The burden of disease and injury in Iran 2003. *Popul Health Metr,* 15;7:9.

16. Torgerson PR, Schweiger A, Deplazes P, et al B (2008). Alveolar echinococcosis: from a deadly disease to a well-controlled infection. Relative survival and economic analysis in Switzerland over the last 35 years. *J Hepatol,* 49:72-77.

17. Murray CJ, Lopez AD, Mathers CD, Stein C (2001). The Global Burden of Disease 2000 project: aims, methods and data sources. *Geneva: World Health Organization,* 36:1-57.

18. Harandi MF, Budke CM, Rostami S (2012). The monetary burden of cystic echinococcosis in Iran. *PLoS Negl Trop Dis,* 6:e1915.

19. Coussens AK, Wilkinson RJ, Nikolayevskyy V, et al (2013). Ethnic variation in inflammatory profile in tuberculosis. *PLoS Pathog,* 9:e1003468.

20. Lopez AD, Mathers CD, Ezzati M, Jamison DT, Murray CJ (2006). *Global burden of disease and risk factors.* ed. The World Bank.

21. Mangen M-JJ, Plass D, Havelaar AH, et al (2013). The pathogen-and incidence-based DALY approach: an appropriated methodology for estimating the burden of infectious diseases. *PLoS One,* 8:e79740.

22. Ao TT, Feasey NA, Gordon MA, Keddy KH, Angulo FJ, Crump JA (2015). Global burden of invasive nontyphoidal Salmonella disease, 2010. *Emerg Infect Dis,* 21:941.

23. Torgerson PR, Devleesschauwer B, Praet N, et al (2015). World Health Organization estimates of the global and regional disease burden of 11 foodborne parasitic diseases, 2010: a data synthesis. *PLoS Med,* 12:e1001920.

24. WHO (2001). The World Health Report 2001: Mental health: new understanding, new hope.

25. Wang Q, Yang L, Wang Y, et al (2020). Disease burden of echinococcosis in Tibetan communities—A significant public health issue in an underdeveloped region of western China. *Acta Trop,* 203:105283.

26. Budke CM, Jiamin Q, Zinsstag J, Qian W, Torgerson PR (2004). Use of disability adjusted life years in the estimation of the disease burden of echinococcosis for a high endemic region of the Tibetan plateau. *Am J Trop Med Hyg,* 71:56-64.

27. Counotte MJ, Minbaeva G, Usubalieva J, Abdykerimov K, Torgerson PR (2016). The burden of Zoonoses in Kyrgyzstan: a systematic review. *PLoS Negl Trop Dis,* 10:e0004831.

28. Torgerson PR, de Silva NR, Fèvre EM, et al (2014). The global burden of foodborne parasitic diseases: an update. *Trends Parasitol,* 30:20-26.

29. Devleesschauwer B, Ale A, Torgerson P, et al (2014). The burden of parasitic zoonoses in Nepal: a systematic review. *PLoS Negl Trop Dis,* 8:e2634.

30. Zhang MY, Wu WP, Guan YY, Wang LY, Hou YY, Feng Y (2018). Analysis on disease burden of hydatid disease in China. *Chinese J Parasitol Parasitic Dis,* 36:15.

31. Modiano D, Petrarca V, Sirima B, Nebie I, Diallo D, Esposito F, Coluzzi M (1996). Different response to Plasmodium falciparum malaria in west African sympatric ethnic groups. *Proc Natl Acad Sci U S A,* 93:13206-13211.

32. Franco MP, Mulder M, Gilman RH, Smits HL (2007). Human brucellosis. *The Lancet Infect Dis,* 7:775-786.

33. Nazirov F (2002). Echinococcosis in Uzbekistan: types of problems and methods to improve treatment. *Med J Uzbekistan,* 2:2-5.

34. Anand S, Hanson K (1997). Disability-adjusted life years: a critical review. *J Health Econ,* 16:685-702.

35. Bern C, Maguire JH, Alvar J (2008). Complexities of assessing the disease burden attributable to leishmaniasis. *PLoS Negl Trop Dis,* 2:e313.

36. Canning D (2006). Priority setting and the 'neglected'tropical diseases. *Trans R Soc Trop Med Hyg,* 100:499-504.

37. Grosse SD, Lollar DJ, Campbell VA, Chamie M (2009). Disability and disability-adjusted life years: not the same. *Public Health Rep,* 124:197-202.

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