

# The pattern of cystic echinococcosis in children in an endemic area in Morocco

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**Abstract** Cystic echinococcosis is a parasitic infection of major public health concern. The infection is generally acquired during childhood and the disease incubation period can last many years. The occurrence and characteristics of the disease in children were not well studied. The present study aimed to explore the pattern and features of cystic echinococcosis in children in rural and urban environments in Morocco. A total of 338 children diagnosed and treated for cystic echinococcosis were investigated. The trend of the infection, risk and exposure factors and the distribution of the anatomic locations of cysts were studied. A non-uniform decrease in proportions of infected children was found. Children from rural environs had significantly

higher infection rates than children from urban environs ( $p < 0.001$ ). Males were significantly more infected than females. Children aged 7–11 years were the most affected. For cysts locations, single organ involvement was found in 94.4% of the children versus 5.6% with multi-organ localization. Despite control program, active transmission of echinococcosis still occurred in children and remains a major public health problem. The infection in younger population may have some features that need to be considered in the prevention and control programs in endemic areas.

**Keywords** Children · Cystic echinococcosis · Cyst location · Environment · Risk factors

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## Introduction

Human cystic echinococcosis (CE) or hydatidosis is a common zoonotic parasitic disease of major public health concern in many countries, especially in endemic areas (Eckert and Deplazes 2004). Human infection by CE occurs by unintentionally ingesting *Echinococcus granulosus* eggs shed in faeces of infected carnivores. The main routes of infection are ingestion of contaminated water or not well-washed ground-growing vegetables and fruits that could be contaminated with eggs of *E. granulosus*, or direct hand-to-mouth faecal transmission which is especially common in children (Craig and Larrieu 2006). There are reports of an increasing number of CE cases in several countries of central Asia, so it is becoming an emerging or re-emerging disease (Torgerson et al. 2006; Torgerson 2013). CE has been included within the group of neglected tropical diseases for which epidemiological surveys are

recommended as part of an effective control approach (Da Silva 2010).

Human CE may affect all ages. Infection occurs mostly at an early age, during childhood or adolescence, and the incubation period is very extensive and may reach many years (Sundarka et al. 2000). CE Prevention measures and control programmes have been implemented in many endemic countries including Morocco. However, studies and published reports on the pattern of CE involvement in children in endemic areas are rare. Therefore, the current study was designed to determine the occurrence, risk and exposure factors of CE in children living in rural and urban environments diagnosed and treated for CE Marrakesh, Morocco.

## Patients and methods

The present survey involved 338 children under the age of 18 years who were diagnosed by imaging techniques (e.g. ultrasonography, computed tomography, X-rays) and operated (predominantly surgically) for CE during a 7-year period (2010–2016) at three major referral governmental hospitals in Marrakesh (Morocco): Ibn Zohr Hospital, Ibn Tofail Hospital and Child and Mother Hospital (University Hospital Centre). These hospitals provide medical care for many areas of the country and are the main consultation centres for patients from several primary healthcare centres. A report containing all the details regarding each CE case was established and completed. In addition to the socio-geographic status (rural versus urban origin) of the children, information on their age, gender, location of the cyst(s), date of admission and diagnosis technique was also determined. For analytical purposes the children with cystic echinococcosis have been divided in three age groups as follows: 2–6 years; 7–11 years; 8–17 years. The trend of the infection during the study period was explored. The anatomic locations of cysts in children were investigated and the distribution according to environment type was determined. Descriptive statistical analysis was performed to assess frequency of CE cases, mean numbers, ranges and annual incidence rate. The Chi square test was used for the comparison of the occurrence frequencies distribution and trends. All data were analyzed using SPSS software (version 17), with a  $p$  value  $< 0.05$  was considered significant.

## Results

Over the study period a total number of 338 cases ( $< 18$  years) have been diagnosed and treated for cystic echinococcosis; ranging from 40 cases in 2016 to 65 cases

in 2010 with an average of 48.3 cases per year (Fig. 1). An overall, but non-uniform, decreasing trend in the number of CE cases was recorded over time, resulting in a decline in the estimated annual incidence rate from 5.3/100,000 to 1.7/100,000 in the region over the study period. The calculated mean annual incidence rate was 3.4/100 000.

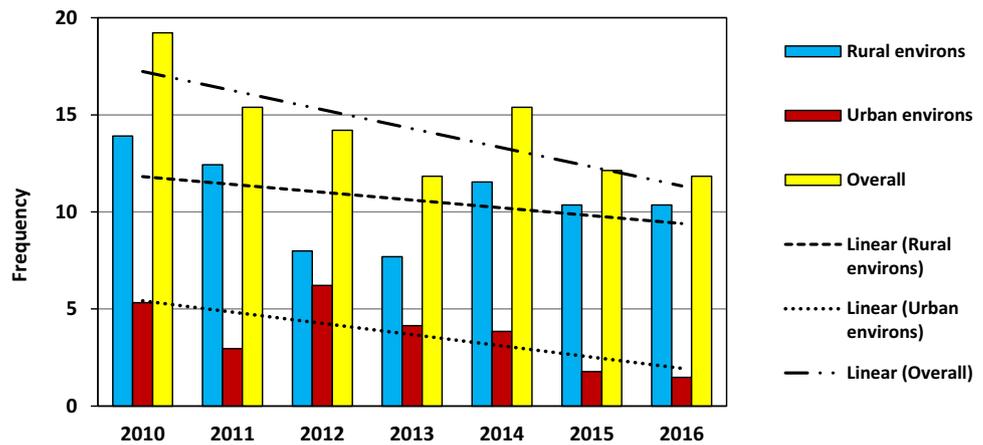
The distribution of the investigated group by environment type (rural versus urban), gender and age is presented in Table 1. Of the 338 children diagnosed and treated for CE, 74.3% (251 cases) were located in rural environs while 25.7% (87 cases) resided in urban environs. The observed difference was statistically significant ( $p < 0.001$ ). For the gender factor, the CE frequency in boys was significantly higher than in girls with respective rates of 57.1% (193 cases) and 42.9% (145 cases). The male to female infection ratio was 1.33:1, and the variation is mainly marked in rural environment ( $p < 0.05$ ). Both males and females hailed from rural environment were more likely to be infected than those of urban areas and the recorded difference was statistically significant ( $p < 0.001$ ).

With regards to children's age, it ranged from 2 to 17 years with a median age of 10 years. The distribution of CE cases by age-groups revealed that 74% of them were between 7 and 17 years old versus 26% of 2–6 years (Table 1). Most of the affected children were 7–11 years old, accounting for 40.8%. Statistical comparisons showed that significantly more children of the age group 7–11 were infected compared with younger age group 2–6 years ( $p < 0.05$ ). For the three defined age groups, the highest rates of CE were recorded among children living in rural environs compared with those who lived in urban environs ( $p < 0.001$ ).

The anatomic location of cysts was investigated (Table 2). Single organ involvement was observed in 319 patients (94.4%). Most of the children had cysts located in the liver than in lungs with respective numbers of 169 cases (50%) and 143 cases (42.3%). The liver: lung infection ratio was 1.18:1. However, the recorded difference was not statistically significant ( $p > 0.05$ ). Uncommon cyst locations (other than the liver and lung) were scarcely detected (7 cases, 2.1%); four children (1.2%) had cysts located in the brain; two cases of peritoneal locations (0.6%) and a single subcutaneous dorsal location (0.3%). No recurrences have been noted. Multiple organ involvement was rare. Only 5.6% of the children (19 cases) had cysts in liver and lung simultaneously, and single to multiple organ involvement ratio was of 16.79:1.

Comparisons of the anatomic distribution of cysts by children's environment revealed a similar pattern of cyst distribution in both children living in rural and urban areas (Table 2). In addition both of single organ involvement (liver or lung cysts) and combined locations were mostly identified in males and in the age group 7–11 years.

**Fig. 1** Trends of cystic echinococcosis cases (< 18 years) diagnosed and treated between 2010 and 2016



**Table 1** Distribution of the diagnosed and treated children for cystic echinococcosis by environment type, gender and age

	Rural environment n (%)	Urban environment n (%)	Total n (%)
Cases	251 (74.3)	87 (25.7)	338 (100.0)
Gender			
Males	143 (42.3)	50 (14.8)	193 (57.1)
Females	108 (32.0)	37 (10.9)	145 (42.9)
Age groups (years)			
2–6	71 (21.0)	17 (5.0)	88 (26.0)
7–11	99 (29.3)	39 (11.5)	138 (40.8)
12–17	81 (24.0)	31 (9.2)	112 (33.2)

**Table 2** The anatomic location of cysts in children diagnosed and treated for cystic echinococcosis, living in rural and urban environments

Cyst location	Rural environment n (%)	Urban environment n (%)	Total n (%)
Single organ			
Liver	126 (50.2)	43 (47)	169 (50.0)
Lung	105 (41.8)	38 (43)	143 (42.3)
Other organs	5 (2.0)	2 (2.3)	7 (2.1)
Sub-total	236 (94)	83 (95)	319 (94.4)
Multiple organ			
Liver-lung	15 (6.0)	4 (2.6)	19 (5.6)
Overall	251 (100)	87 (100)	338 (100.0)

**Discussion**

Human CE is one of the most geographically widespread zoonotic infections, representing a major public health problem in many endemic countries (Eckert and Deplazes 2004; Budke et al. 2006; Jairajpuri et al. 2012). CE may affect all age groups. Infection occurs mostly during

childhood although years may elapse before symptoms are seen. As a result, especially for organs with thicker consistency localizations, the disease diagnosis and treatment is often much later (Jordanova et al. 2015). Surveys are required to determine risk factors, trends and features of the disease in younger population. The current study revealed a declining tendency in the incidence of CE through the 7-year study period. This is in agreement with the findings of Pierangeli et al. (2007) reporting a non-uniform decrease in CE annual incidence from 22.1/100,000 to 6.2/100,000 in children, over a 10-year period, in a region undergoing a control program in Argentina.

The study results showed that children living in rural zones are more likely to be highly affected than those residing in urban environs, with proportions reaching 74.3% versus 25.7%, respectively. However, the high occurrence of CE in urban areas may be overestimated due to cases originated from surrounding peri-urban areas or having a repeated travel history to rural high-risk zones where favourable conditions for CE transmission are encountered.

In the Moroccan context, the significant predisposing factors responsible for CE transmission in rural areas are the permanent relationship between dogs and domestic livestock, and the close contact between children and dogs. Furthermore a lack of awareness among children and parents about the infection factors, the poor compliance with the hygienic practices, low standard of living and lack of sanitation may result in high infection levels (Vlad et al. 2013; Grimason et al. 2014; Chakarova et al. 2015). A previous survey carried out by El Berbri et al. (2015) revealed that in 39% of the investigated households owned dogs have access to the family home (including the kitchen); 79% of the surveyed group were unaware of the role dogs play in CE transmission. Also 61.2% of children did not use to wash their hands after petting or playing with dogs. Additionally, home slaughtering mainly during cultural and religious celebrations is common in Morocco, and

dogs are often fed on the discarded offal due to people's unawareness of the risks associated with this practice. The illegal slaughtering practice, inadequate infrastructure of abattoirs in rural areas, as well as the neglectful practices of slaughterhouse workers through discarding affected organs to free roaming dogs tend to maintain CE transmission (El Berbri et al. 2015; Jordanova et al. 2015). Moreover, most of the inhabitants are involved in farming works. Due to the close proximity of agricultural workers' houses to fields, children are often observed playing in and around the fields coming into contact with contaminated soil. Such exposure presents an additional risk to children of becoming infected due to their imprudence whilst playing and their pica by eating unusual substances followed by direct hand-to-mouth transfer (Amahmid and Bouhoum 2005). The children of agriculturists are usually involved in farming works such as field irrigation and shepherding. These practices involve working with hands in the egg-contaminated soil and plants, as well as contact with livestock and dogs.

Water supplies contamination by the eggs of *Echinococcus*, irrigation of vegetables with contaminated water and soiling of vegetables by dogs may be a significant source of infection in rural areas (Dowling et al. 2000). Additionally, in rural environment, conditions such as relative humidity and temperature may be favourable to parasite eggs survival, remaining infective for months (Veit et al. 1995). So, in rural environment, many conditions and practices may increase the potential for CE transmission and infection (Chebli et al. 2017). The findings of the current study are in concordance with the reports of Hezarjaribi et al. (2017), in Iran, and Vlad et al. (2013) who found that CE was twice more frequent in children of rural areas as compared to urban regions in Romania. In urban environment, children infection may be attributed to soil contamination in parks and school gardens with *Echinococcus* eggs shed by numerous stray dogs and to closer contact between children and dogs (Dopchiz et al. 2009; Grosso et al. 2012; Jordanova et al. 2015).

For the gender factor, significantly more males than females were infected. Boys and girls living in rural areas are significantly more exposed to CE than those living in urban environment. Behavioural activities and habits increasing exposure to infection sources are the main reasons for these gender differences. Males are more exposed to CE because, in Morocco, boys tend to spend more time outdoors and are more involved in farming works in rural areas than girls (Amahmid and Bouhoum 2005). Boys, more so than girls, are considered to be closer canine companions and may be more at risk to potential zoonotic infection through the faecal-oral route during the course of feeding, working or playing with their dogs. The results of this study corroborate those previously reported in other

paediatric studies quoting that boys are more likely to be infected than girls, with male to female ratios ranging from 1.3:1 to 2.1:1 (Wang et al. 2001; Durakbasa et al. 2006; Fahimzad et al. 2015). In contrast, a predominance of CE in females has been reported (Pierangeli et al. 2007; Sanaei Dashti et al. 2017).

With respect to age, children from 7 to 11 and 12 to 17 years were significantly more infected compared with those aged between 2 and 7 years, with marked significant levels in children living in rural environment. The changes occurring in children's behaviour, practices and life activities with age tend to increase the chance of being infected with CE (Morrongiello et al. 2001). In addition, the clinical signs of CE may require years to appear due to slow disease progress with age (ECDC 2014). The obtained results are consistent with those reporting that pre-adolescent years (8–12 years) accounted for the most frequent age and CE is rarely identified in patients under 7 years of age (Mirshemirani et al. 2011; Vlad et al. 2013).

Based on the study results in relation to cyst location, single organ involvement was predominant in the investigated children. Liver and lung were the most commonly affected organs with a slight, but non-significant, higher rate for liver cysts. The greater involvement of the liver and lung occurs because the larvae released from ingested eggs penetrate the intestinal wall and move through the circulatory system into liver then lung. Therefore, the liver acts as the primary filter in the human body and the lung is often thought to be the second filter before the larva can reach other organs (Alghoury et al. 2010; Kohansal et al. 2015). Comparative conclusions were made in previous surveys (Ok et al. 2007; Djuricic et al. 2010). However, most authors found lung cysts to be more frequently identified than liver cyst locations in children (Durakbasa et al. 2006; Pierangeli et al. 2007; Mirshemirani et al. 2011). This matter was attributed to rapid growth of cysts in lung tissue due to its lower resistance compared to liver (Tantawy 2010). Detection is also associated with the noisier and earlier symptoms of lung cysts reported in children (e.g. cough, chest pain) encouraging earlier parental intervention to seek medical help (Chaouachi et al. 1989). For uncommon anatomic cyst locations and combined liver and lung cysts, similar findings were quoted by Djuricic et al. (2010). Contrary to children, it has been reported that liver to lung infection ratios were significantly higher in adults (Todorov and Boeva 2000; Vlad et al. 2013). Several previous surveys reported that unusual locations are significantly rare in children compared with adults, while combined liver and lung involvement are more frequently identified in younger population than adults (Tsaroucha, et al. 2005; Talaiezhadeh and Maraghi 2006; Jairajpuri et al. 2012).

The distribution of the anatomic cyst location by children's origin showed that the solitary and multi-organ involvement in children living in rural and urban environments is identical.

## Conclusion

Cystic echinococcosis is an endemic disease in Morocco and inter-sectoral program for the disease prevention and control was implemented targeting humans, dogs and livestock populations as well as standardization of slaughterhouses. The undertaken study allowed an assessment to be made of CE distribution in children as well as factors and conditions increasing children exposure risk and infection transmission. The occurrence of the infection in children indicates active transmission of the parasitic infection from the main sources to children especially in rural areas. Consequently, the designed control program should be fully implemented by the involved departments in order to achieve effective CE prevention and control, taking into account possible differences in CE pattern and features in children and adults (Talaiezadeh and Maraghi 2006; Djuricic et al. 2010). Among the integrated control tools, organising awareness campaigns to raise the whole community (adults and children) knowledge and change attitudes and behaviours may be an important component to interrupt CE transmission. In order to gain better insight into the problem with this zoonosis, additional epidemiological studies in children and adults, other intermediate hosts and definitive hosts have to be accomplished in CE endemic areas.

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## Compliance with ethical standards

**Conflict of interest** The authors declare that there is no conflict of interest.

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