



RESEARCH ARTICLE

Determination of risk factors and patient profile in human hydatidosis

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İnsanlarda hidatidozis'te risk faktörlerinin ve hasta profilinin belirlenmesi

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Öz

Amaç: Bu çalışma, insan hidatidozis'inde (kistik ekinokokkozis) hasta profilini belirlemek ve risk faktörlerini ortaya koymak için Afyonkarahisar'da gerçekleştirilmiştir.

Gereç ve Yöntem: Hidatidozis tanısı konulan ve cerrahi tedavi alan 166 tane hastanın hastane kayıtları çıkartılmış ve kendilerine 30 sorudan oluşan bir anket uygulanmıştır.

Bulgular: Hidatidozis insidansının, kadınlarda (%53) erkeklerden (%47) daha yüksek olduğu görülmüştür. Kistler en yüksek oranda karaciğer (%53) ve akciğerlerde (%40,4), en düşük oranda ise beyinde(%1,2) görülmüştür. Enfeksiyon en yüksek 50-59 (19,9) ve en düşük 0-9 (%2, 4) yaş gruplarında görülmüştür. En yüksek oran ilk-orta (%49,1) öğrenim grubunda ve en düşük oran da (%0,6) lisans üstü eğitim grubunda bulunmuştur. En yüksek oranda yıllık geliri 10.000 TL'den düşük gelirli grupta (%48,8) ve en düşük olarak da 50.000 TL ve üzeri gelir grubunda (%0,6) görüldüğü saptanmıştır. İnsidans kırsalda ikamet edenlerde (%62) şehirde ikamet edenlerden (%38) daha yüksek oranda görülmüştür. Hidatidozis'in köpek sahibi olanlarda daha yüksek (%54,8), olmayanlarda daha düşük (%45,2) oranlarda görüldüğü saptanmıştır.

Öneri: Hidatidozis, özellikle endemik olduğu ülkelerde büyük sosyo-ekonomik etkileri olan çok önemli bir halk sağlığı problemi olarak kabul edilmektedir. Bu araştırma ile Türkiye'de halk sağlığını tehdit eden önemli bir paraziter zoonoz olan hidatidozis'te hasta profili, demografisi ve risk faktörleri kapsamlı olarak ortaya konulmuştur.

Anahtar kelimeler: Hidatidozis, insan, risk faktörleri, hasta profili

Abstract

Aim: This study was carried out in Afyonkarahisar to determine the patient profile and to reveal risk factors in human hydatidosis (cystic echinococcosis) out in Afyonkarahisar region, Turkey.

Materials and Methods: The hospital records of 166 patients diagnosed with hydatidosis and treated surgically were evaluated. The patients were subjected to a questionnaire consisting of 30 questions.

Results: The incidence of hydatidosis was found to be higher in women (53%) than in men (47%). Cysts showed the highest proportion in the liver (53%) and in the lungs (40.4%) and lowest in the brain (1.2%). Infection was exclusively concentrated between 50 and 59 (19.9%) year old patients and the lowest was in the 0-9 (2, 4%) age group. The rate was 49.1% in the elementary-secondary education group and 0.6% in the post graduate studies group. There was a close association between the incidence and patient's income. Patients had lower income (less than 10.000 TL) showed higher incidence (48.8%) whereas those having higher income (50.000 TL and over) had the lowest incidence rate (0.6%). Incidence was higher in rural residents (62%) than in urban residents (38%). Hydatidosis was found to be higher in dog owners (54.8%) than in non-dog owners (45.2%).

Conclusion: Hydatidosis is considered a very important public health problem with major socio-economic impacts in countries where it is particularly endemic. Patient profiles, demographics and risk factors are extensively presented in hydatidosis, an important parasitic zoonosis that threatens public health in Turkey in this study.

Keywords: Hydatidosis, human, risk factors, patient profiles





Introduction

Hydatidosis (cystic echinococcosis) is a zoonotic metacestode infection caused by the larval stage of taeniid cestode *Echinococcus granulosus* (Batsch, 1786). Hydatidosis, which has a high public health significance is prevalent worldwide in humans and domestic animals (Thompson 1995; Pedro and Schantz 2009; Mandal and Mandal 2012). Typical cystic lesions can occur in many different parts of the body, especially the liver and lungs. In addition to economic losses caused in livestock breeding, which has a critical impact in human nutrition, it also threatens human health. Income losses caused by high surgical and medical treatment costs, hospital costs, diminished quality of life, deaths, loss of work force are a heavy burden for the country's economies, while causing a number of health problems that can end in death in humans. When the losses in cases that cannot be diagnosed and treated are taken into consideration, hydatidosis is a zoonosis of high socio-economic importance in terms of public health. (Eckert et al 2001; McManus et al 2003; Torgerson and Budke 2003; Budke et al 2006; Craig et al 2007). It has been estimated that the annual global effect of hydatidosis on human health and the livestock breeding industry is about 3 billion US dollars (Budke et al 2006).

The larvae of *E. granulosus* develop as discrete and single (unilocular) cysts. For this reason in humans it is encountered in a milder and less treatable form. Furthermore, multiple or large cysts can cause anaphylactic reactions as a result of permanent damage or rupture in the organs where they are located. In farm animal breeding, animals are often slaughtered before cysts display clinical signs. Long-lived animals are clinically symptomatic. Economic losses incur when internal organs are destroyed as a result of cysts detected in post mortem meat inspection as well as meat and milk yield loss (Eckert et al 2001; Thompson 2001; Eckert and Deplazes 2004).

Hydatidosis can occur in people of all ages and sexes. In humans, hydatid cysts are found in the brain and spleen, kidneys, bones, heart, other organs and tissues, although they are highly localized in the liver and lungs. The ratio of liver infections to lung infections is expressed as 2.5:1 (Eckert and Deplazes 2004) and 65%: 25% (Schwabe 1986). When the eggs are ingested orally, primary cysts are formed in organs and tissues and secondary cysts are formed in case they rupture. Approximately 40-80% of hydatid cyst patients present cysts in a single organ. Generally, unilocular cysts can cause pathological disorders and various clinical findings in the relevant organs depending on their placement within the organs, their size and complications such as rupture of the cyst. Hydatid cysts rarely cause death in cases of vital organ involvement, surgical complications and rupture. (Zapatero et al 1989; Eckert et al 2001; Thompson and McManus 2001; Eckert and Deplazes 2004).

Hydatidosis is a cosmopolitan metacestode infection that commonly found in humans and animals worldwide. The infection

is endemic in some Mediterranean countries, the Middle East, South America, Africa, Southeast Asia and Oceania. Infection is encountered more frequently in developing countries and rural areas, but also in the population outside the endemic areas as a result of increased travel and international trade. Worldwide, 2-3 million cases of hydatidosis are reported annually in humans (Craig et al 2007).

The incidence of hydatidosis in Bulgaria, where hydatidosis is a major public health problem, was reported to be 6.5/100.000 during 1950-1962, 2.2/100.000 during 1971-1982 and 3/100.000 during 1983-1995 after the control campaign in 1960 (Todorov and Boeva 1999). In Greece, retrospective surveys indicated that the incidence was 9.77/100.000 between 1969-1975 and that the incidence of surgical cases was 7.9/100.000 (Karpathios et al 1985) and 12.7/100.000 (Papadopoulos 1985) between 1981-1983 and that this ratio had decreased after the eradication program was carried out in 1984. An incidence of 3.8/100.000 has been reported for the western parts of Romania (Calma et al 2011) while the annual incidence rate for the south-west and Midwest regions has been reported as 3.3/100.000 (Moldovan et al 2012). The incidence rate for Italy has been reported as 1.92/100.000, however the incidence rate for the infection in Sardinia where it is endemic was reported as 20/100.000 during 1975-1980 and 6.62/100.000 during 2001-2005 period (Conchedda et al 2010).

The incidence in humans in Kyrgyzstan has been reported as 5.4/100.000 in 1991 and 18/100.000 in 2000 (Torgerson et al 2003). It is estimated that the incidence in dogs in the former Yugoslav republics may be as high as 65%, and the incidence in humans can also be high while in France it is 4.5-13/100.000, it is 2.2/100.000 in Portugal and 2.5/100.000 in Spain and no very strict control program has been implemented in Malta, it has been reported that a very strict control program was implemented in Malta and no longer seen, that the disease was very common in Cyprus before 1970 and it was minimized through the control program that was implemented between 1971-1985 (Vuitton and Economides 2004). It has been reported that the incidence observed in Uruguay was 20/100.000 during 1962-1974 and 55/100.000 during 1993 (Carmona et al 1998). The incidence in the Central Asian countries in Kazakhstan was between 0.9 and 1.4 during 1997-1994 and 1.4-6.4/100.000 during 1994-2003, 17.8-16.5/100.000 in Uzbekistan, 25/100.000 in Tajikistan and 17/100.000 in Turkmenistan (Torgerson et al 2006). It has been suggested that the incidence of surgery in Peru of South America is 32-127/100.000, 6-20/100.000 in Chile and 1.4-30/100.000 in Argentina (Pedro et al 2006).

According to data from the Ministry of Health, 52.154 patients in Turkey were operated for cystic echinococcosis treatment between 1990 and 2005 which corresponds to an average of 3.257 cases per year while the estimated rate of surgical cases was 0.87 to 6.6/100.000 and the incidence was 0.8-2/100.000 (Altıntaş 2008). A retrospective study conducted in hospitals bet-



ween 2001 and 2005 (Yazar et al 2008) showed that 13.13% of 14.789 cases of surgical hydatidosis (6.30/100.000) had taken place in Marmara region, 16.9% in the Aegean region, 39.58% in Central Anatolia, 5.70% in the Black Sea Region, 6.80% in Eastern Anatolia and 2.7% in Southeast Anatolia. Echinococcosis and hydatidosis in Turkey are quite common in animals. The prevalence in dogs is reported to be between 0.9-44% (Güzel et al 2008), in cattle, sheep and goats it has been reported as 8.96-46.41%, 3.50-70.91% and 1.6-29.8% respectively (Beyhan ve Umur 2011). A number of retrospective studies have been conducted in terms of analysis and determination of risk factors in hydatidosis cases and infection in various parts of the world. In Italy's Sardinia where hydatidosis is endemic, the incidence of infection was determined as 6.62/100.000 in a retrospective survey covering 2001-2005, the number in rural areas increased to 14 and the ratio of male patients versus female patients was 1.36, an increased risk of infection with increased age was reported, the liver had been affected in 72% of all cases and lung cysts were more common in men than in women (Conchedda et al 2010). In a study of 61 hydatidosis patients (12 males, 49 females) in Iraq's Hilla, the prevalence of infection was highest in the 34-45 age group (32%) and lowest in the 15-24 age group (8%), 82% of the patients lived in the rural area and 87% of them had animals or had contact with animals (Al-Yasari et al

2013). According to a survey of the residents of Lima's suburbs carried out to determine the risk factors in Peru, it was manifested that infected dog owners, those who fed their dogs raw organs, those who frequently had contact with dogs in childhood and those who use and drink water without boiling had a higher incidence of the disease (Moro et al 2008).

According to the results of a survey conducted in China's Ningxia Hui autonomic region, hydatidosis is encountered at a higher rate in those who are over 30 years of age and have had a dog for at least 5 years and consume unhealthy water without boiling (Yang et al 2006). A questionnaire carried out with 144 shepherds, 119 breeders, 25 slaughterhouse workers and 80 hydatidosis patients in Jordan revealed that they had very little knowledge of how people became infected while 1.2% of the patients who underwent surgery constituted patients with hydatidosis (Nasrieh et al 2003). In a study conducted in Aydın, Turkey, hydatidosis was reported mostly in the liver, followed by the lungs, and more frequently in women; according to the results of the survey, 44.01% of the respondents had dogs, 84.79% of them did not use anthelmintics on their dogs, 6.40% threw cystic organs offal into the garbage while 84.22% had no knowledge of hidatidosis (Ertabaklar 2012).

Table 1. Demographic Characteristics of Hydatidosis Patients

Age group	Patient numbers n (%)	Gender		Residence		Dog owner			Educational Status			Income status (Annual TL)				
		M (%)	F (%)	Urban (%)	Rural (%)	Y (%)	N (%)	E-S (%)	H (%)	U (%)	PG/D (%)	<10.000	10.000-20.000	20.000-30.000	30.000-40.000	≥50.000
0-9	4 (2.4)	2 (2.6)	2 (2.3)	1 (1.6)	3 (2.9)	3 (3.3)	1 (1.3)	1 (1.3)	0 (0.0)	0 (0.0)	0 (0.0)	4 (4.9)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
10-19	22 (13.3)	8 (10.3)	14 (15.9)	6 (9.5)	16 (15.5)	10 (11.0)	12 (16.0)	7 (8.8)	10 (23.8)	5 (12.8)	0 (0.0)	20 (24.7)	2 (3.7)	0 (0.0)	0 (0.0)	0 (0.0)
20-29	31 (18.7)	14 (17.9)	17 (19.3)	6 (9.5)	25 (24.3)	19 (20.9)	12 (16.0)	3 (3.8)	11 (26.2)	16 (40.0)	1 (100)	15 (18.5)	8 (14.8)	6 (24.0)	2 (40.0)	0 (0.0)
30-39	26 (15.7)	16 (20.5)	10 (11.4)	8 (12.7)	18 (17.5)	11 (12.1)	15 (20.0)	10 (12.5)	7 (16.7)	9 (22.5)	0 (0.0)	4 (4.9)	11 (20.4)	10 (40.0)	1 (20.0)	0 (0.0)
40-49	24 (14.5)	10 (12.8)	14 (15.9)	8 (12.7)	16 (15.5)	13 (14.3)	11 (14.7)	12 (15.0)	5 (11.9)	7 (17.5)	0 (0.0)	9 (11.1)	7 (13.0)	5 (20.5)	2 (40.0)	1 (100)
50-59	33 (19.9)	19 (24.4)	14 (15.9)	14 (22.2)	19 (18.4)	16 (17.6)	17 (22.7)	23 (28.8)	8 (19.0)	2 (5.0)	0 (0.0)	14 (17.3)	15 (27.8)	4 (16.0)	0 (0.0)	0 (0.0)
60-69	18 (10.8)	5 (6.4)	13 (14.8)	13 (20.6)	5 (4.9)	12 (13.2)	6 (8.0)	16 (20.0)	1 (2.4)	1 (2.5)	0 (0.0)	12 (14.8)	6 (11.0)	0 (0.0)	0 (0.0)	0 (0.0)
70≥	8 (4.8)	4 (5.1)	4 (4.5)	7 (11.1)	1 (1.0)	7 (7.7)	1 (1.3)	8 (10.0)	0 (0.0)	0 (0.0)	0 (0.0)	3 (3.7)	5 (9.3)	0 (0.0)	0 (0.0)	0 (0.0)
Total	166 (%)	78 (46.99)	88 (53.01)	63 (37.95)	103 (62.05)	91 (54.82)	75 (45.18)	80 (48.20)	42 (25.30)	40 (24.10)	1 (0.60)	81 (48.80)	54 (32.53)	25 (15.06)	5 (3.01)	1 (0.60)

n: Number of patients, M: Male patient, F: Female patient, E-S: Graduate of Elementary and secondary school, H: High school graduate, U: University graduate, PG/D: Post graduate studies/Doctorate





The profile, demographic information and risk factors in terms of infection in hydatidosis patients will be extensively presented with this study comprising a 3-year period covering 2012-2014.

Materials and Methods

The patient records of a university hospital between 2012-2014 were examined. Medical, percutaneous and surgical treatment records were obtained for patients who were pre-diagnosed for hydatidosis by means of radiological examinations (Ultrasound, MRI and CT), serological and allergic tests. The organ localizations of cysts, hospitalization times and patient profile (sex, age etc.) were also obtained. The demographic information of the patients (place of residence, education status, livelihoods, annual income levels, social security status, receiving protective care services, personal awareness about the infection, perception about hygiene and sanitation, owning a dog, etc.) was gathered by a questionnaire (contained 30 questions) applied to the patients. In data analysis, patient profiles and demographic status were compared and possible risk factors were defined. IBM SPSS Statistics for Windows, Version 21.0. (IBM Corp.; Armonk, New York, USA). Pearson Chi-Square (Monte Carlo) test was used for the analysis of the data.

Results

The prevalence of hydatidosis according to patient age groups, demographic characteristics such as the number of patients, gender, residence (rural, urban), dog ownership and income status is given in Table 1.

The incidence of hydatidosis was 47% in men and 53% in women indicating no statistically significant difference between the sexes ($p > 0.05$). Hydatidosis was highest in the liver (53%) and lowest in the brain (1.2%). The prevalence in lungs was determined to be (40.4%) while coexistence in the liver and lungs was 5.4%. The difference between single organ (94.6%) cases and multiple organ (5.4%) cases in terms of organ location was statistically significant ($p < 0.001$).

According to age groups and organ location, infection was highest in the 50-59 age group (19.9%) and lowest in the 0-9 age group (2.4%). There is no statistical difference between the age groups in terms of the incidence of infection in organ locations ($p > 0.005$).

According to the education level, hydatidosis was highest in the elementary-secondary education group (49.1%) and the lowest was in the post-graduate studies group (0.6%). There was no significant association between organ location and educational status ($p > 0.05$).

According to income status, the highest rate of hydatidosis was observed in the group with an annual income less than 10.000 TL (48.8%) and the lowest in the group with an income of

50.000 TL and more (0.6%). There was no difference between the income groups in terms of organ location ($p > 0.05$)

When the residence and age were taken into consideration, the infection was higher in rural areas (62%) than in urban areas (38%) indicating statistically significant difference ($p < 0.05$). The highest rate of hydatidosis was observed in the 50-59 age group living in rural and urban areas.

When dog ownership and the frequency of hydatidosis is compared, the ratio was higher (54.8%) among dog owners and lower (45.2%) in non-dog owners and the difference was found to be significant ($p < 0.05$).

In the 7.2% of the patients with hydatidosis had a recurrence after treatment while 92% had no recurrence. No difference was observed in organ location in recurrent cases ($p > 0.05$). Seventy five (82.4%) out of the 91 dog owner patients who were dog owners did not medicate their dogs against cestode infections with any anthelmintics while 16 dog owners (17.6%) used anthelmintics and 82 dog owners (90.1%) fed raw organs and offal to their animals while 9 (9.9%) did not.

Hundred and seven of the patients (64.5%) reported that they consumed green salads outside (i.e. restaurants) while 59 patients (35.5%) claimed they did not. The number of patients who reported that they washed raw vegetable and fruit before consumption was 154 (92.8%) while 12 patients (7.2%) answered that they did not wash fruit and vegetables. Thirty four patients (20.5%) consumed untreated water (lakes and running waters) while 132 (79.5%) did not.

The number of patients who had other family members afflicted with hydatidosis other than themselves was 27 (16.3%) while the number of those without such an affiliation was 139 (83.7%). Fifty six patients (33.7%) were aware of the disease before they were diagnosed while 110 (66.3%) had no knowledge on hydatidosis. The number of patients informed about transmission and protection after being diagnosed with the illness was 164 (98.8%) while 2 (1.2%) indicated that they had not been informed.

Patients ($n=153$) had stray dogs in their environment while 13 patients had no contact the dog in their milieu. Patients 157 (94.6%) washed their hands after handling dogs while 9 (5.4%) did not. Patients 93 (56%) had been instructed about hygiene in the family environment or in school while 73 (44%) had not been instructed in this regard. All the patients in this study had been treated by conservative surgery. Except for one of the patients, they all had social security.

Discussion

Hydatidosis is considered to be a very important public health problem with major socio-economic impacts, especially in co-



ountries where it is endemic. A series of studies have been carried out in various countries (Nasrieh et al 2003; Li et al 2005; Yang et al 2006; Moro et al 2008; Conchedda et al 2010; Ahmadi and Badi 2011; Moldovan et al 2012; Vahedi and Vahedi 2012; Al-Yasari et al 2013; Banda 2013; Singh et al 2013; Li et al 2015) and in Turkey (Ertabaklar et al 2012; Akalın et al 2014) in order to determine the patient profile and risk factors of hydatidosis, which is an important zoonosis threatening public health in Turkey as well as farm animals. This thesis study has been carried out in order to determine possible differences in the patient profiles of infected patients as well as risk factors due to the socio-economic and cultural structures of the countries.

The executed studies indicate that there are differences in the distribution of hydatidosis according to gender. Only one survey (Conchedda et al 2010) indicated that the proportion of men with hydatidosis was higher while most researchers (Carmona et al 1998; Li et al 2005; Calma et al 2011; Ahmadi and Badi 2011; Moldovan et al 2012; Ertabaklar et al 2012; Vahedi and Vahedi 2012; Akalın et al 2014) indicated a higher incidence in women. The result of this study (53% for women and 47% for men) supports this.

Some researchers have indicated that the risk of hydatidosis is higher in middle aged individuals. 30-39 and 40-49 (Carmona et al 1998), 23-30 and 30-40 (Vahedi and Vahedi 2012), 35-44 (Al-Yasari et al 2013), 21-40 (Ahmadi and Badi 2011), 50-54 (Calma et al 2004), 50-59 (Moldovan et al 2012) and Akalın et al 2014 reported increased hydatidosis risk in the 30-39 age group while some researchers (Carmona et al 1998; Conchedda et al 2010) reported that the risk of hydatidosis increased with age. In this study, infection was found to be highest in the age range of 50-59 (19.9%).

Considering the educational status of hydatidosis patients reveals that the proportion of illiterate and elementary school graduates is higher (Akalın et al 2014; Wang et al 2014; Li et al 2015). The results of this study support this data. The highest rate (49.1%) was found in elementary-secondary school graduates.

It has been reported that single organ manifestation of the cysts in patients is more common than multiple organ manifestation and that cysts are mainly located in the liver (Carmona et al 1998; Torgerson et al 2003; Conchedda et al 2010; Calma et al 2011; Ahmadi and Badi 2011; Moldovan et al 2012). The results of this study corroborate these findings with the ratio of single organ manifestation of cysts (94.6%) and multiple organ manifestation (5.4%) which are mainly in the liver (53%).

In most of the studies reported that the rate of infection was higher in rural areas (Conchedda et al 2010; Calma et al 2011; Al-Yasari et al 2013; Akalın et al 2014) while in two studies it was higher in the city population (Ahmadi and Badi 2011; Moldovan et al 2012). In this study, the infection rate (62%) in rural areas

was found to be higher.

It is reported that the incidence of infection is higher among dog owners or those who are in close contact with dogs (Torgerson et al 2003; Nasrieh et al 2003; Li et al 2005; Moro et al 2008; Al-Yasari et al 2013; Singh et al 2013; Wang et al 2014). The result of this study supports this statement (54.8%).

It is argued that those who do not treat their dogs with anthelmintics against cestode infections, those who feed raw materials to their dogs and those who consume greens without washing (Torgerson et al 2003) are more susceptible to infection. The results of this study concur with these results. The percentage which does not treat their dogs with anthelmintics was 82.4% while the proportion of those who feed their dog with uncooked organs and offal was 90.1%.

Conclusion

Echinococcus granulosus for which 10 different genotypes (G1-G10) have been currently defined with the development of molecular techniques and which has been identified as a complex of species/genotypes as a result of molecular genetic studies based on mitochondrial DNA analyses, continues to be threat to animal health, the livestock breeding industry as well as public health. From this perspective, it is highly probable that this topic will continue to be a focal point for relevant researchers.

Continuous epidemiological updates are being made on the subject with various studies. A limited amount of research is available on the risk factors of hydatidosis which has a high incidence in livestock and humans especially in some parts of the world and in developing countries. There are, of course, differences in the socio-economic, cultural and educational circumstances of the countries concerned. In Turkey, only a few parameters such as gender, age and location have been studied in addition to studies on the incidence of infection with retrospective research in humans. In this study, the risk profile, demographic information and risk factors of hydatidosis patients in Afyonkarahisar during the 3-year period covering 2012-2014 have been examined extensively. This study will shed light for researchers in the prevention and control of helminthic zoonosis hydatidosis, which is a threat to public health, animal health and the relevant industry.

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Ethics board approval

This study has been approved by the ethics committee of Afyon Kocatepe University Clinical Research Board.





Patient informed consent

Verbal consent has been obtained from patients whose hospital records were used in the study and who participated in the survey.

Conflict of interest

Authors have not reported a conflict of interest.

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