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Original Article

Evaluation of characteristics of the patients who undergo surgical treatment for lung hydatid cyst

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ABSTRACT

Background: Hydatid cyst is a public health problem, particularly where it is endemic. It is a benign disease and usually asymptomatic unless the symptoms and complications develop. Effective treatment is important because this benign disease can cause considerable mortality and morbidity. The aim of this study was to describe the epidemiological and clinical characteristics of HC, localization (bilateral lungs or multiple organ involvement) and perforation characteristics of HC.

Materials and Methods: 160 patients were operated on for pulmonary hydatid disease for about approximately 12 years of time period. Patients were evaluated according to their age, gender, distribution of cysts in the lung, extra-pulmonary organ involvements, presence or absence of perforations, and animal contact stories.

Results: Number of the cysts, the cyst diameter, gender, location of the cysts and the liver involvement were found unrelated with the perforation status of the cyst. But, there is a significant difference between the localization of the cysts in the lung and the involvement of the liver. The rates of presence of liver cysts in the case of unilateral right, unilateral left and bilateral cysts in the lung were found as 13%, 25% and 50% respectively.

Conclusions: Hydatid cyst is a public health problem and elimination is possible with rigorous work. In case of bilateral pulmonary cysts and the multiple number of cysts, the likelihood of a cyst in the liver is increased. It was concluded that there was no relationship between the presence of cysts in the liver and the absence of perforation in the lung cysts. However, it should be kept in mind in terms of the possible complications that may occur at high perforations in patients who received preoperative albendazole therapy.

Key Words: albendazole monohydrochloride, cystic echinococcosis, hydatid cyst, perforation of cyst

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Introduction

Turkey is one of the countries where hydatid cyst (HC) is considered endemic [1]. According to WHO, worldwide, 1 million people are living with these diseases at any one time. Annual costs are estimated to be 3 billion US \$. The prevention programs are directed to the definitive hosts like dogs and lambs for cleaning them from the parasites. The WHO Informal Working Group on Echinococcosis (WHO-IWGE) continues to guide and set priority areas [2].

The disease is caused by the larvae in the metacystod stage of the *Echinococcus granulosus*. Four type of echinococcus have been defined. *Echinococcus granulosus* is the most common infestation type in human [3]. *Echinococcus multilocularis* is less common and causes alveolar echinococcosis. *E. vogeli* and *E. oligarthus* causes polycystic echinococcosis and they are endemic in South America.

Humans acquire the infection by oral ingestion of parasite eggs that contaminated water and food. Parasites in the mesenteric circulation reach the liver through the portal system. They cannot pass through the sinusoids and that's why this disease most commonly occur in the liver (55-70%). The second most frequent site is lungs (18-35%) [1]. Parasites can reach the lung by hepatic vein, vena cava inferior, right lung, pulmonary arteries road and also reach by transdiaphragmatic transition, lymphatically or by portocaval anastomosis. Both liver and lung can be affected in about 5-13% of all cases [4]. It's reported in different studies that it could be placed in almost all organs.

HC is a benign disease and usually asymptomatic unless the symptoms and complications develop. Clinical course varies according to the organ in which it is placed, effects on adjacent tissue, its superinfections and hypersensitivity reaction that can cause anaphylaxis. If lung HC is ruptured towards the bronchus, the patient can describe membrane and clear water expectoration and hemoptysis. Effective treatment is important because this benign disease can cause considerable mortality and morbidity [5].

The diagnosis of HC depends on clinical findings, imaging methods and serology. There is no common consensus on the HC therapy strategy, medical treatment (albendazole), surgery and percutaneous drainage.

The definitive treatment of lung hydatid disease is surgery. In cases where there is a possibility of cyst spread, it is recommended to use albendazole and mebendazole as postoperative preservatives [6].

The aim of this study was to describe the epidemiological and clinical characteristics of HC, localization (bilateral lungs or multiple organ involvement) and perforation characteristics of HC.

Materials and Methods

A total of 254 patients with pulmonary hydatid cyst diagnosed between January 2006 to March 2018, in Thoracic Surgery Clinic in our institution. Patients who were thought to be hydatid cysts in preoperative radiological examinations but could not be confirmed with postoperative pathology results were excluded from the study. In addition, patients who did not have evaluation for extrapulmonary organ involvement were also excluded from the study. Finally, 160 patients were studied retrospectively. An illuminated affirmation form of the permission or prohibition of the use of their medical information for medical research purposes is signed by all patients admitted to our clinic. Patients were evaluated according to their age, gender, distribution of cysts in the lung, extra-pulmonary organ involvements, presence or absence of perforations, and animal contact stories. This study has been approved by Institutional Review Board of our institution.

Statistical Analysis

SPSS for Mac Version 20.00 (SPSS Inc., Chicago, IL, USA) was used for the statistical analysis of the data obtained at the end of the study. Continuous variables were expressed as mean \pm standard deviation (SS), while categorical variables were as number and percentage (%). The continuous variables were assessed by the Kolmogorov-Smirnov test for normal distribution. Chi-square test for intermittent variables and Student-t test for continuous variables were used for comparison of groups. Statistical significance was accepted as $p < 0.05$.

Results

160 patients were operated on for pulmonary hydatid disease for about approximately 12 years of time period. Age and gender characteristics of the patients, lung localization of the cysts, extra-pulmonary organ involvements, perforation status of the cysts and animal contact story of the patients were evaluated.

146 patients were male (91.25%), whereas the remaining 14 (8.75%) were female. Their mean age was 24.99 ± 9.6 (20-66). Mean age in females was higher than males (41.14 ± 15.8 versus 23.44 ± 7.2) ($p < 0.001$).

When compared to the number of cases according to years, it is seen that in 2009 and 2011 there were more cases, the number of cases has gradually decreased in recent years (Table 1).

Table 1. Number of cases by years.

Year	Number of cases (n)
2006	12
2007	16
2008	17
2009	20
2010	19
2011	19
2012	14
2013	12
2014	12
2015	9
2016	6
2017	1
2018	3
Total	160

The mean number of cysts in all patients was 1.51 (min: 1, max: 9) and the mean cyst diameter was 6.42 cm (min: 1cm, max: 25cm). Both the number of the cysts and the cyst diameter were not statistically significant in relation with perforation status of the cyst (respectively, $p = 0.150$, $p = 0.843$). Also, there was no statistically significant relationship between gender and the perforation status of the cyst ($p = 0.392$).

62 (38.8%) of the patients had a history of animal contact, while 98 (61.2%) of them did not had. The history of animal contact was significantly higher in males than females (41.1% versus 14.3%) ($p = 0.049$).

There was a similar result between existence of extrapulmonary organ cyst and the history of animal contact which is not statically significant ($p = 0.984$). In 14 of 62 patients (22.6%) with the positive animal contact history had extrapulmonary organ cyst, while in 22 of 98 patients (22.4%) with no animal contact history had extrapulmonary organ cyst. On the other hand, cyst perforation ratio was higher in patients with no history of animal contact (44.9% versus 30%), but this is not statistically significant ($p = 0.063$).

When the distribution of the cysts according to lobes were evaluated, it was found that cysts were most frequently located unilateral 88.2% and in the right lung 51.2%. In 11.2% of all patients, cysts were located bilaterally.

There was no statistically significant difference between the localizations of the cysts and the perforation status (Table 2).

Table 2. Distrubution of cysts in the lung.

	Lober Localization (n)	Perforded / Intact	p
Right n,% (82, 51.2)	Upper lobe (n=22)	11/11	0.839
	Middle lobe (n=19)	8/11	0.547
	Lower lobe (n=45)	23/22	0.551
Left n,% (60, 37.5)	Upper lobe (n=25)	7/18	0.906
	Lingula (n=1)	0/1	0.534
	Lower lobe (n=34)	12/22	0.171
Bilateral n,% (18, 11.2)	Upper lobe (n=8)	2/6	0.502
	Middle lobe-Lingula (n=5)	1/4	0.457
	Lower lobe (n=17)	6/11	0.467

37 patients had extrapulmonary involvement. Except one case with spleen and heart involvement 35 (22.2%) patients had liver involvement.

The presence or absence of liver involvement did not affect the perforation status of the cysts ($p = 0.374$).

There was no significant difference between liver involvement and cyst diameter ($p = 0.938$), however, as the number of cysts in the lung increased, the rate of liver involvement increased ($p = 0.009$).

The rates of presence of liver cysts in the case of unilateral right, unilateral left and bilateral cysts in the lung were found as 13%, 25% and 50% respectively.

Discussion

Cystic hydatidosis continues to be a public health issue for endemic countries. According to health statistics Republic of Turkey, in 2001 annual incidence has been reported as 4.9 in 100000 [7].

This parasitosis is more common in areas where the livestock are existence and the work of men in this business is more usual in Turkey. In parallel with this, 91.25% of our patients were males.

Hydatid cyst is usually acquired in childhood, but most are diagnosed at age 30-40 [8]. Tor et al. reported that 31% of the patients were younger than 20 years of

age in their pulmonary hydatid cysts series which containing 288 patients [9]. Only adult patients were operated on in our clinic so in our study the mean age was found 24.99 (20-66 years).

In this study the number of annual cases decreased in an approximately 10-year period. While the hydatid cyst prevalence is closely related to poor sanitation conditions, this can be explained by the improvement of these conditions over the years and by the successful treatment of animals.

Perforation is the most common complication of pulmonary hydatid cysts [10]. In previous studies, perforation was thought to be related to the with the demographic characteristics of the patient, number of cysts, intra-cystic pressure and localization of the cysts [11,12].

The relationship of intrathoracic pressure with cyst perforation has also been investigated, previously. It is thought that the factors that increase intrathoracic pressures such as coughing and trauma may be responsible for perforation [12,13].

Intra-cystic pressure increases as the diameter of the cyst increases. So, even small increases in cyst size can lead to perforation by increasing intra-cystic pressure. It should be kept in mind that diagnostic invasive procedures may also be responsible for perforation [14].

There is no consensus to support the association of number and diameter of cysts with perforation of any of them. Kocer et al reported no significant relationship between increasing the diameter of the cyst reduces the risk of perforation [15]. Burgos et al claimed that increasing the diameter of the cyst reduces the risk of perforation [16]. We could not determine the relationship between number and diameter of cysts with perforation in our study. However, especially during childhood, as cysts grows, the pressure inside the cyst increases due to the low volume of the thorax. Therefore the rates of cyst perforation in children are reported higher [17].

The number of literatures comparing cyst perforation with cyst localization is low. The rate of perforation in our study was 40%. This ratio is compatible with the rate ranging from %24.7 to 61% in the literature. In our study, the most frequently placed lobe of the cysts was the left lower lobe while the perforation was most

frequently observed in the right lower lobe. This data is in contrast to Onal et al, which reports that perforation is most commonly seen in the right middle lobe cysts in their series of 134 cases [12].

People become infected by digesting parasite eggs in contaminated food, water or soil or by direct contact with the host. It is reasonable that the disease is more frequent in rural areas where there is more animal contact with humans. In our study, 38.8% of patients had an animal contact story. Compared to females there was higher ratio of animal contact in males (41.1% vs 14.3%). This can be explained by the fact that men are more engaged in animal livestock in rural areas.

There is no agreed evidence that more cysts are formed or more organs are involved in case of animal contact history. There was no statistically significant difference between extrapulmonary cysts existence and animal contact story in our study. Also, we found that cyst perforation is not related to animal contact history.

Albendazole treatment is accepted to be effective and safe in hydatid cysts of the liver. It is reported that preoperative use of albendazole reduces the viability of the cyst while the postoperative use of it decreases the recurrence [18].

In adults, lung is the second most common localization after liver. Most chest surgeons face with lung cyst hydatid on the chest x-ray that took for control while receiving albendazole treatment for liver cyst hydatid. Albendazole is able to penetrate into hydatid cysts, and active against protoscolices of *Echinococcus granulosus* in vitro cultures. Drug effect by interfering with glucose absorption in the parasite wall and ultimately cell autolysis comes to the forehead. This may result in cyst perforation for the lung. In our study, in 18 patients (48.6%) cysts were perforated from 37 patients who underwent preoperative medical treatment. This high perforation ratio is compatible with the literature [5,14].

In conclusion, hydatid cyst is a public health problem and elimination is possible with rigorous work. Compatible with the literature data, it was seen that animal contact was more frequent in male patients. The location of the cyst in the lung does not affect the risk of perforation. In case of bilateral pulmonary cysts and the multiple number of cysts, the likelihood of a cyst in the liver is increased. It was concluded that there was no re-

relationship between the presence of cysts in the liver and the absence of perforation in the lung cysts. However, it should be kept in mind in terms of the possible complications that may occur at high perforations in patients who received preoperative albendazole therapy.

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Declaration of conflicting interests

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References

1. Sachar S, Goyal S, Goyal S, Sangwan S. Uncommon locations and presentations of hydatid cyst. *Ann Med Heal Sci Res* 2014; 4: 447–52.
2. World Health Organisation, Echinococcosis. <http://www.who.int/news-room/fact-sheets/detail/echinococcosis>.
3. Brunetti E, Kern P, Vuitton DA. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. *Acta Trop* 2010; 114: 1–16.
4. Safioleas M, Misiakos E, Manti C. Surgical treatment for splenic hydatidosis. *World J Surg* 1997; 21: 374–8.
5. Kuzucu A, Soysal Ö, Özgel M, Yologlu S. Complicated hydatid cysts of the lung: Clinical and therapeutic issues. *Ann Thorac Surg* 2004; 77: 1200–4.
6. Sanlı M, Tuncozgun B, Elbeyli L. Pulmonary hydatid disease and surgical therapy. *Turkish J Thorac Cardiovasc Surg* 2000; 8: 703–5.
7. Ministry of Health (TR), Research Planning and Coordination Council: Health Statistics 2001 <https://dosyamerkez.saglik.gov.tr/Eklenti/23522,2001-yili10pdf.pdf?0> Turkish.
8. Arinc S, Kosif A, Ertugrul M, Arpag H, Alpay L, Ünal Ö et al. Evaluation of pulmonary hydatid cyst cases. *Int J Surg* 2009; 7: 192–5.
9. Tor M, Atasalihi A, Altuntas N, Sulu E, Senol T, Kir A et al. Review of cases with cystic hydatid lung disease in a tertiary referral hospital located in an endemic region: A 10 years' experience. *Respiration* 2000; 67: 539–42.
10. Yekeler E, Karaarslan K, Yazıcıoğlu A, Kaya SKN. Lobectomy for pulmonary hydatid cyst. *Turk J Med Sci* 2013; 43: 1024–9.
11. Yuksel M, Kir A, Ercan S, Batirel HF, Baysungur V. Correlation between sizes and intracystic pressures of hydatid cysts. *Eur J Cardio-thoracic Surg* 1997; 12: 903–6.
12. Onal O, Demir O. The relation between the location and the perforation rate of lung hydatid cysts in children. *Asian J Surg* 2017; 41: 422–6.
13. Leviav S, Weissberg D. Traumatic rupture of hydatid cysts. *Canad J Surg* 1996; 39: 293–6.
14. Cobanoglu U, Sayir F, Şchitoğlu A, Bilici S, Melek M. Therapeutic strategies for complications secondary to hydatid cyst rupture. *Int J Clin Exp Med* 2011; 4: 220–6.
15. Kocer B, Gulbahar G, Han S, Durukan E, Dural K, Sakinci U. An analysis of clinical features of pulmonary giant hydatid cyst in adult population. *Am J Surg* 2009; 197: 177–81.
16. Burgos L, Baquerizo A, Munoz Wi de Aretxabala X, Solar C, Fonseca L. Experience in the surgical treatment of 331 patients with pulmonary hydatidosis. *J Thorac Cardiovasc Surg* 1991; 102: 427–30.
17. Cangır A, Sahin E, Enon S. Surgical treatment of pulmonary hydatid cysts in children. *J Pediatr Surg* 2001; 36: 917–20.
18. Arif SH, Shams-ul-Bari, Wani NA, Zargar SA, Wani MA, Tabassum R, et al. Albendazole as an adjuvant to the standard surgical management of hydatid cyst liver. *Int J Surg* 2008; 6: 448–51.