

Research Article

A study of the frequency of medical and surgical treatment of hydatid cyst in the patients of Arak city, Iran

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Abstract: Hydatidosis is one of the most important parasitic diseases worldwide, and it is caused by the larval stage of *Echinococcus granulosus*. This disease is endemic in Iran and can cause large losses of life and property. A few studies have been done in this area in Arak city, Iran. This study aims to investigate the prevalence of medical and surgical treatment of patients with hydatid cyst in hospitals of Arak city to promote the management of patients. This study is a descriptive study in which the cases are examined and the characteristics of patients with hydatid cyst and their relevant treatment method in Vali ASR and Amir al-Moemenin Hospitals between 2009-2013 have been evaluated. Of 70 patients, 41 (58.6%) were women and 29 (41.4%) were men and the highest incidence was in the third decade of age (20%). Most patients belonged to the rural population (54.3%). The organs with highest involvement were liver (62.9%) and lung (27.1%) respectively, and the involvement of other organs was (7.14%). The average size of the cysts was 33±85 mm. 64 patients (91.42%) received combined treatment, and 6 patients (8.57%) only received medical treatment. The highest and most effective treatment of choice in the patients with hydatid cyst is the combined surgery and pharmaceutical procedure, and medication therapy before and after surgery, reduces relapses.

Keywords: Hydatidosis, *Echinococcus granulosus*, Pharmaceutical and surgical procedure

INTRODUCTION

Hydatid cyst is an infectious disease shared between humans and animals that is spreading around the world and is it has been familiar to man since ancient times, so that Hippocrates also referred to it [1]. The disease is caused by infestation of humans and animals with the larval stage of *Echinococcus granulosus*. Humans become infested by swallowing the eggs of *E. granulosus*.

Dogs and sheep are definitive and intermediate host of this parasite, respectively. However, man becomes involved by having infested vegetables and food. The higher risk of this disease is due to the fact that it can involve critical and sensitive members of body such as liver and lung, etc. [2]. The most commonly affected organ is the liver, and lungs, central nervous system, eyes, etc. may also be involved as well.

This disease is more prevalent in the areas with much presence of animals such as sheep and canines like Iran, Turkey, Mediterranean Countries, Middle East, South America, New Zealand and Australia [3]. The disease is usually asymptomatic and clinical manifestations are different depending on the location of the cyst. The incidence of hydatid cyst in endemic areas of the world has been reported 10 per hundred

thousand people [4]. In Iran, the highest and lowest geographical spread of the disease is in Khorasan and Hormozgan provinces, respectively. However, statistics and studies in recent decades indicate that the highest prevalence of the disease is fields near mountainous areas and its geographical distribution perfectly matches the side-mountainous climate zones of the country [5]. This disease does not have belongs to a certain age and affects all age groups. Contamination rate in both sexes has been reported equally. Hydatid cyst may appear anywhere on the body but it involves liver in 70% of cases and in 15 to 20% of cases involves the lungs [6].

Many cases are asymptomatic. When the cyst appears in the liver, symptoms are mainly abdominal pain and sensitivity, the mass presence in the abdomen or large diffusion in liver. And when the cyst is in the lungs, it appears mainly as cough, shortness of breath, transient chest pain or hemoptysis [6].

Nowadays, the treatment of single hydatid cyst is surgery, but some patients with multiple lesions in various organs or due to certain physical conditions cannot be a candidate for surgery. On the other hand, the results are not always successful surgery and in some cases it has been with secondary diffusions or local recurrence. In these cases, drug therapies are used

[7]. Given that one percent of surgeries annually in Iran are related to Hydatidose and most of aged patients cannot be treated and get afflicted with secondary cyst and impose a high cost to families and state both in terms of sanitary and economic considerations, some effective strategies should be designed [8].

Also, in some cases, patients with hydatid cyst are not treated completely, and given that the disease is aboriginal in Iran and can cause economic damages to health system and insurer companies and also due to the existence of medical and surgical problems for patients, employing a combination of medical and surgical methods for the treatment of hydatid cyst is of utmost significance. As many medical and surgical procedures have been used in Iran and the world in the past, detailed information on the statistics of the disease in the Arak city is not available.

The current study mainly aims to investigate the frequency of medical and surgical treatment procedures for the patients with hydatid cyst referred to Arak hospitals between 2009-2013, in order to suggest scientific recommendations consistent with the disease information in other countries and to present some applicable strategies for an effective and valuable planning in the treatment of the patients with hydatid cyst.

MATERIALS AND METHODS

The present study is of a descriptive type in which some cases are examined, and characteristics of patients with hydatid cysts and their treatment method in Vali Asr & Amir al-Moemenin Hospitals since 2009 to 2013 is analyzed. The study population is composed of all the patients hospitalized in different parts of the above mentioned hospitals, which the term “hydatid cyst” was diagnosed as the initial diagnosis in their file. First of all, a checklist of the all the variables under study including age, gender, residency place, organ affected, the number of cysts, cyst size, treatment and relapse documents were prepared. Then, by referring to the archives of the hospitals, the records of the patients undergoing medical or surgical treatment of hydatid cyst on the determined date were examined and the required information extracted.

For a more accurate investigation of the treatment procedure, the patients were evaluated both in terms of initial treatment and post-relapse treatment separately. Also, the patients were examined in different age decades and the size of the cysts was divided into some groups. The results in the frequency tables have been reported in the form of the number and percentage. For quantitative indicators, the mean and standard deviation are used. For qualitative indicators, two-dimensional tables as the number and percentage were used. The software used was SPSS version 16.

As the study data are extracted from the files, so the lack of the record of some variables in patients’ files and unavailability of the patients’ treatment results are some of the limitations of the present study.

RESULTS

Similar to other diseases, age distribution of the patients with hydatid cyst is varied at different age groups due to the different behavior of each age group to its environment. In the present study, the maximum prevalence in terms of age was related to the third decade with the frequency of 20%, then second and fourth decade with 18.16% and finally the first decade with 1.4%. The sexual distribution of hydatid cyst is different in both genders, and this is a natural state due to their relationship with environment. Of 70 patients, 41 (58.6%) were female and 29 were male (41.4%).

In terms of residence place of the patients with hydatid cyst, the most of the patients lived in rural areas with the frequency of 54.3% and the remainder lived in civil places. Of 70 patients under study, in 44 (62.9%) the liver was involved. The liver involvement was observed in 19 cases (27.1%), the concurrent involvement of liver and lung was observed in two cases (2.9%), and other organs’ involvement was observed in five cases (7.14%) including brain (2 cases, 2.9%), plevis (2 cases, 2.9%) and cardiac (one case, 1.4%). The frequency rate of the cysts is presented in Table 1.

Table 1: The frequency rate of the cysts in the patients with hydatid cyst

Number of Cyst	Frequency	Percent
1	48	68.6
2	11	15.7
3	2	2.9
4	2	2.9
5	1	1.4
6	1	1.4
>7	5	7.1

The mean size of the cysts was 33±85mm. The frequency rate of the cysts’ size is presented in Table 2.

Table 2: The frequency rate of the cysts in the patients with hydatid cyst

Size of Cyst (mm)	Frequency	Percent
1-25	1	1.4
26-50	7	10
51-75	21	30
76-100	19	27.1
101-125	13	18.6
126-150	4	5.7
151-175	4	5.7
176-200	1	1.4

Some patients were treated for the first time and some had an unsuccessful treatment in the past. Of

the patients who have received pharmaceutical treatment, treatment of seven patients was not successful and they were treated again. Finally, 64 patients (91.42%) were treated at once, and only 6 patients (8.57%) were treated by a pharmaceutical intervention. It should be mentioned that all the patients who underwent surgical treatment, after and before surgery received pharmaceutical treatment. The surgical method of all the patients was laparotomy and the medication prescribed for all patients was Albendazole. Of 70 patients, 8 (11.4%), their disease recurred which underwent re-treatment. Among the 8 patients who relapsed, 7 patients (87.5%) were received previously only drug treatment, and 1 patient (12.5%) received a combination of treatments once before. Also to obtain further information, we did other tests which the results are as follows: The average size of cyst was in the patients who had combined therapy was higher, but there was no significant correlation between them. The average age in pharmaceutical was higher, but there was not a significant relationship between them.

The number of the patients living in civil areas was higher but there was not a significant relationship between them. There was not a significant relationship between the treatment procedure and the variables including age, sex, involved member and the number cyst.

DISCUSSION

Markazi Province is one of the most important centers in the country for raising sheep and goats. And as the prevalence of hydatid cyst is directly associated with agriculture and husbandry, they considered as the areas prone to hydatidose. Therefore, it is necessary to take necessary actions and to train people on this issue. In the present study, the obtained data almost adapts to the resource and research conducted, and in case of any difference the main reason is mentioned. The age of patients in difference resource is the second and third decade in terms of hydatose prevalence. In this study, of 70 patients hospitalized in Ark hospitals, the third decade accounted for the maximum frequency flowed by second and fourth decades, respectively. In the study conducted by Sarkari *et al.*; in Yasouj since 1995 to 2005 and Kamhawi in Jordan, the maximum frequency of disease was reported in the third decade [9, 10]. In a study by Salehi *et al.*; in 2009 to 2011 in Khorasan province with the maximum rate of hydatose prevalence in Iran, the most patients has the age range of 30 to 41 [11], which the fourth decade in our study had the maximum frequency. Given that the duration of disease in hydatid cyst is long, the manifestation of clinical symptoms in the middle ages in this disease explains the high rate of incidence of hydatid cyst in the third decade of life. The sexual distribution of hydatid cyst in both sexes is different and this is a natural state due to their relationship with environment. In the present study, of 70 hydatid cyst, 41 cases (58.6%) were female and 29 cases (41.4%) were male.

The main reason of affliction with hydatid cyst in the women has not mentioned in any references, while it can be due to high contact of women with livestock and infectious cause of parasite egg. Also, the studies conducted by Salehi *et al.*; since 1999 to 2012 in Khorasan province, Davud Abadi *et al.*; in Kashan in 1997 to 2005 and Jacob since 1967-1982 in Canada reported that the maximum rate of affliction with this disease was related to women [11-13]. In most of studies [14-17], there is not so much difference between rural and civil regions. Contact to livestock in rural area and consumption of infested vegetables can raise the likelihood of infestation in both groups. In our study, most of the infested people were rural people with the frequency of 54.3% and people living in civil areas had the frequency of 47.7%. This result is consistent with the study conducted by Salehi *et al.*; [11] in Khorasan. These problems results from low level of personal and public hygiene in villages, profession requirements of villagers and their close relationship with dogs of flocks. According to the references, the most common organ in the granulosis infestation is liver (65-75%) followed by lung (20-30%). Given that the hydatid eggs swallowed pass from intestinal wall and enter into blood circulation, they can be replaced in each tissue or organ. However they are mainly found in liver and lungs. 80-90% of human hydatids are evolved in these two organs, and mostly in the liver. In a study conducted on 70 patients with hydatid cysts, 44 patients (62.9%) had hepatic involvement. Other studies have also confirmed the higher involvement of liver in liver hydatidosis. Also, 19 cases (27.1%) had pulmonary involvement. In most studies in Iran and around the world, liver and then lung has been the most affected of all the organs of the body. 2 cases (2.9%) had a concurrent involvement of liver and lungs. In a study by Davud Abadi *et al.*; in Kashan in 1997-2005, 68% of the patients were afflicted with hepatic hydatid cyst and 30% were with pulmonary hydatid cyst [12]. In a study by Salehi *et al.*; (2009-2012) in Khorasan province, the maximum rate of involvement was related to liver and lung [11].

The main cause of the hepatic involvement is the pathogenic mechanism of the hydatid cyst. In addition, 4.1% had concurrent involvement of lung and liver, which was consistent with the results of Aredkani in Kerman, Sarkari *et al.* in Yasouj, Shiryazdi in Yazd and Ghafari in Babol [9, 15, 18, 19]. Primary brain involvement is very rare, so that in endemic areas includes about 0.2-2% of all cases, which can be caused by the blood release of larvae, and may lead to seizures or symptoms of the icp [20]. However, in this study, we had 2 cases (2.9%) of cerebral involvement. Cardiac involvement can lead to mechanical rupture and tamponade [21, 22]. Also, we had one case of cardiac infection (1.4%) and one cardiac involvement of the heart (2.9%). In terms of the frequency of the cyst, the highest frequency was one single cyst in 48 patients

(68.6%) followed by the highest number of two cysts is (15.7%), and multiple cysts were 5 cases (7.1%). In terms of the frequency of cyst size, the group 75-51 mm, the maximum frequency, 21 cases (30%), then the group 76-100 mm, 19 cases (27.1%), and then group 101-125mm, 13 cases (18.6%), and minimum frequency is less than 25 mm, 1 case (1.4%) and over 176 mm, 1 case (1.4%). The mean size of the cysts was 85 ± 33 mm.

In the study conducted by Dogru and colleagues in Ankara, Turkey, in 2004 the average size of the cysts was 59 ± 38 mm [23] that less than the figure obtained in our study. Traditionally, surgery has been the first definitive treatment. Albendazole is used to treat simultaneously. Its administration begins a few days before surgery and continues for several weeks thereafter [24]. People who for whatever reason do not want surgery, the elderly, and patients with serious illness at the same time, and those who have a small-size cyst can only get medical treatment [25]. Medical treatment with albendazole alone for 12 weeks to 6 months resulted in cure in approximately 30% of the cases and the resultant improvement in 50% of the cases [26]. In 70 patients with hydatid cysts in Arak hospitals, some patients were treated for the first time and some had a previous unsuccessful treatment. Of these 70 patients, 57 (81.42%) underwent combined surgical and medical treatment, which one patient underwent combined treatment at the next visit. And 13 patients (18.57%) received the primary drug therapy, which the primary drug treatment of seven patients was not successful and they were treated at their last hospitalization. Finally, 64 patients (91.42%) received combined treatment, and only six patients received only drug therapy. No patients had surgical treatment without medication. The surgical procedure of all patients was Laparotomy and surgical patients prescribed drug in all of them was albendazole. Of 70 patients, 8 (11.4%) had a relapse which were re-treated. Of 8 patients who had a relapse, 7 patients (87.5%) had already received medication therapy, and one patient (12.5%) received combined treatment once, which showed that the maximum relapse has happened in medication procedure.

In a study by Amary *et al.*; in Jordan (2002) [27], Aref *et al.*; (2001-2003) in Kashmir [28], Iels *et al.*; (2002) in London, Bildick *et al.*; (2007) [30] and Actan *et al.*; (1996) [3], the most effective and prevalent treatment method was the combined treatment procedure and medication therapy before and after surgery played an auxiliary role along with surgery and had minimized relapse rate. The studies in which dominant treatment was not combined method are much lower. For example, in the study of Tatar *et al.*; in 1996-2001 in Turkey, surgical treatment was reported in 7 cases (41.1%), the combined medical and surgical treatment was reported in 3 cases (17.6%) and drug therapy was reported alone in 7 cases (41.1%), and it

was concluded that surgery should be the treatment of choice, but drug therapy should be considered in uncomplicated or unresectable cases [32].

Jamshidi *et al.*; (2007-2008) in Hormozgan followed 9 patients with multiple cysts treated with albendazole and praziquantel for 3 months and then reported complete clinical recovery in 7 cases (77.7%) and relative clinical recovery in two cases (22.2%) [33]. This rate was lower than the recurrence rate of the patients in our study, but due to lack of follow-up of the patients in our study, the studies cannot be compared with each other.

Also to obtain further information, other tests were performed, which the results are as follows: The average size of cyst in the patients who underwent combined treatment was higher, while there was not a significant correlation between them.

In a study in Ankara, Turkey in 2004, there was a significant correlation between the size of the cyst and the treatment results. The study suggested that children with uncomplicated pulmonary hydatid cyst with size less than 5 cm should have a close follow-up treatment with an experimental drug [23]. In a study by Todoro *et al.* in response to chemotherapy, the very important factor was the size as well, that was more effective in smaller and younger cysts [34]. The mean age was higher in drug treatment but there was no significant correlation between them. The number of the patients living in civil areas in the combined procedure was much more; however there was no significant correlation between them. There was no significant correlation between treatment procedure and age, sex, organ involvement and the number of the cyst as well.

CONCLUSION

Based on our study and the previous research that some of them was presented, it was concluded that the most effective alternative treatment in patients with hydatid cyst is combined procedure in which surgery is used as the basis for treatment, and medication therapy after and before operation is used as complementary treatment along with surgery and can reduce recurrence. Also, medication therapy is not solely sufficient and a high percent of it leads to failure and relapse. Also, in some cases, the low percentage of patients who have relapsed after adjuvant therapy, had not completed already their course of medical treatment, or operating conditions was not sterile and post-operative care was not very well done. Although the average size of cyst in the patients who had received medication therapy was lower, there was not a significant association between them, which this can be due to small size of the study sample and it requires further studies. Regarding the variables including age, sex, organ involvement, and number of cysts in the choice of treatment, this study did not find a significant relationship, but cannot rule

out the impact of them, and it is necessary to further investigate a larger population of patients.

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