



SCIENTIFIC ARTICLE

Our clinical experience and follow-up results in hydatid cyst cases: a review of 393 patients from a single center



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Abstract

Background and objectives: Hydatid cyst is a zoonotic disease caused by *Echinococcus granulosus*. The aim of our study is to present the clinical features of the patients who were treated for hydatid cyst, determine the interventional techniques and anesthesia methods used and review the occurred complications in detail.

Methods: This study included 393 patients who were followed up and/or treated with the diagnosis of hydatid cyst between January 2013 and November 2018. The patients' data was evaluated retrospectively.

Results: The mean age of the patients was 31.0 ± 17.2 years. Of the patients, 111 (28.4%) had more than one cyst and 36 (9.2%) patients had multi-organ involvement. Six of the patients refused the intervention or was transferred to another hospital. Among the remaining 387 patients, 335 (85.2%) received general anesthesia and intubation, 9 patients (2.3%) received general anesthesia and laryngeal mask airway, 39 patients (9.9%) received sedoanalgesia and 4 patients (1%) received regional anesthesia. Perioperative mortality was developed in one patient. The most common perioperative complication was allergic reaction (1.5%), whereas the most common post-operative complications were atelectasis (3.3%) and biliary fistula (3%). The mean Intensive Care Unit stay (ICU) was 1.9 ± 1.1 days in patients requiring ICU. Recurrence during the 40 ± 17 months follow-up occurred in 8.4% patients.

Conclusions: Anesthesiologists have an important role in the management of hydatid cyst patients. Patients should be evaluated exhaustively in terms of multi-organ involvement and the presence of more than one cyst in the same organ. The type of treatment procedure and the localization of the cysts determine the anesthetic management.

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PALAVRAS-CHAVE

Cisto hidático;
Tipos de anestesia;
Complicações

Nossa experiência clínica e resultados no acompanhamento de casos de cisto hidático**Resumo**

Introdução e objetivos: Cisto hidático é uma zoonose causada por *Echinococcus granulosus*. O objetivo do estudo é apresentar as características clínicas dos pacientes submetidos a tratamento de cisto hidático, determinar as técnicas intervencionistas e os tipos de anestesia utilizados, e revisar as complicações ocorridas, em detalhe.

Método: Este estudo incluiu 393 pacientes que foram acompanhados e/ou tratados com o diagnóstico de cisto hidático, entre Janeiro de 2013 e Novembro de 2018. Os dados dos pacientes foram avaliados retrospectivamente.

Resultados: A idade média dos pacientes foi $31,0 \pm 17,2$ anos. Do total de pacientes, 111 (28,4%) tinham mais de um cisto, e 36 (9,2%) apresentavam comprometimento em vários órgãos. Seis pacientes recusaram a intervenção ou foram transferidos para outro hospital. Dentre os 387 pacientes remanescentes, 335 (85,2%) receberam anestesia geral e intubação, 9 (2,3%) anestesia geral e máscara laríngea, 39 (9,9%) sedação e analgesia, e 4 (1%) anestesia regional. Houve um óbito no período perioperatório. Reação alérgica foi a complicação perioperatória mais comum (1,5%), e no pós-operatório, observou-se mais atelectasia (3,3%) e fístula biliar (3%). O tempo médio de internação na Unidade de Terapia Intensiva foi $1,9 \pm 1,1$ dias para aqueles que necessitaram desses cuidados. Recidiva durante o seguimento de 40 ± 17 meses ocorreu em 8,4% dos pacientes.

Conclusões: Os anestesiológicos têm um papel importante no tratamento dos pacientes com cisto hidático. Os pacientes devem ser avaliados exaustivamente em relação a comprometimento de vários órgãos e presença de mais de um cisto no mesmo órgão. O tipo de procedimento terapêutico e a localização dos cistos determinam a conduta anestésica.

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Introduction

Hydatid cyst disease is a zoonotic disease caused by *Echinococci*, which is more common in populations largely engaged in animal husbandry.¹ It has a global distribution and is considered an endemic disease in many places, such as the Mediterranean, Middle East, Eastern Europe, Australia, South America and African countries. Its prevalence in Turkey is currently lower compared to the past years but the disease remains a considerable problem. The prevalence of hydatid cyst disease ranges from 5 to 40 per 10,000 individuals in our country and it is higher in regions where animal husbandry is common such as the rural areas of Eastern and Southeastern Anatolia.^{2,3}

The disease is more common in young people and it most commonly affects the liver while many other organs can also be involved.⁶ The lung and spleen are the most commonly involved organs after the liver. The heart and the brain are less commonly involved.⁷ However, the growth rate of the cyst can vary depending on the organ. Pulmonary hydatid cyst cases may present symptoms earlier, as the cysts reach larger sizes compared to those in other organs, due to the spongy nature of lung tissue.⁸ Hydatid cysts with slower growth remain asymptomatic for a long time and become symptomatic when they reach sizes enough to compress the surrounding organs and when they cause allergic reactions due to cyst rupture.⁶

There are different treatment options for hydatid cysts including medical treatment, percutaneous treatment and surgical treatment.² The anesthetic method to be used may vary based on the choice of interventional treatment.⁹

Life-threatening complications such as cyst rupture and anaphylaxis may occur during interventional treatment. In addition, Intensive Care Unit (ICU) may be required for some complications that occur during the postoperative period. For all these reasons, anesthesiologists play an important role both during interventional treatment and postoperative patient management.

The aim of this study is to present the clinical features of the patients treated for hydatid cyst at our clinic, to identify the anesthetic methods used for surgery and to review the arising complications in detail.

Methods

In this study, we retrospectively analyzed the data of 393 patients followed up and/or treated with the diagnosis of hydatid cyst in Sanliurfa Mehmet Akif Inan Training and Research Hospital between January 1, 2013 and November 1, 2018. All patients who were diagnosed with hydatid cyst during this period were included in the study. However, the patients who received another diagnosis while being investigated with a preliminary diagnosis of hydatid cyst and in whom the diagnosis of hydatid cyst was excluded during surgery were excluded from the study. Ethics approval for the study was granted by the Ethics Committee of Harran University Faculty of Medicine.

Patient data was collected from the hospital's data system, archived patient charts and surgical records. Demographic data of all patients were recorded. In addition, data including cyst size, localization, multiple

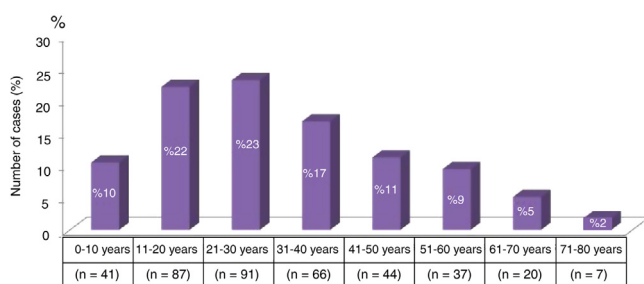


Figure 1 Distribution of hydatid cyst disease by age decades.

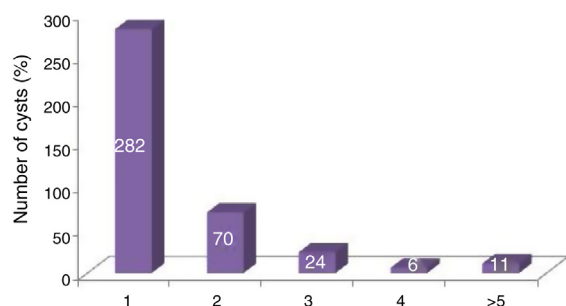


Figure 2 Number of cysts in study patients.

organ involvement, treatment modality, anesthetic method, perioperative/postoperative complications, requirement of postoperative intensive care, intensive care unit stay, and recurrence during follow-up were recorded. The data obtained were recorded on study forms and evaluated.

Statistical analysis

IBM SPSS 24.0 (Statistical Package for the Social Sciences, IBM Corporation, Armonk, NY, USA) was used to analyze data. Continuous variables were expressed as mean \pm standard deviation and categorical variables were presented as numbers (percentages). The conformity of continuous variables to normal distribution was analyzed using the Kolmogorov-Smirnov test. Student *t*-test was used to compare continuous variables and Chi-Square test was used to compare categorical variables. Pearson's correlation coefficient was used for correlation analyses. A *p*-value of <0.05 was considered statistically significant.

Results

Of the 393 patients, 257 (65.4%) were female and 136 (34.6%) were male, and the mean age was 31.0 ± 17.2 years (range: 3–78 years). When the patients were grouped with respect to age decades, the majority of the patients were in the second (22%) and third (23%) decades of life and 72% of the patients were in the first 4 decades of life (Fig. 1). In terms of ethnicity, 344 patients (87.5%) were of Turkish origin, while 49 patients (12.5%) were of Syrian origin living in Turkey.

While 282 patients (71.6%) had only one cyst, 111 (28.4%) patients had more than one cyst (Fig. 2). Three hundred

Table 1 Distribution of cysts by organs.^a

	N° (%)	Percentage (%)
Liver	325	75.2
Lung		
Bilateral	14	3.2
Right	50	11.6
Left	18	4.2
Spleen	10	2.3
Kidney		
Right	7	1.6
Left	2	0.5
Soft tissue	2	0.5
Heart	2	0.5
Omentum	1	0.2
Retroperitoneal	1	0.2

The values are expressed as number and percentage.

^a Patients may have multi-organ involvement.

Table 2 Preoperative patient characteristics.

	N° of patients (n)	Percentage (%)	Mean \pm SD
Multi-organ involvement	36	9.2	
Mean n° of cysts			1.5 \pm 1.0
Cyst size			
<5 cm	37	9.4	
5–10 cm	287	73	
>10 cm	69	17.6	
Preoperative cyst rupture	50	12.7	

The values are expressed as mean \pm SD or number and percentage.

N°, number; cm, centimeter.

and twenty-five patients had hepatic cysts, 82 had pulmonary cysts, 10 had spleen cysts and 9 patients had renal cysts. As for the other less commonly affected organs, two patients had cardiac cysts, and omental and retroperitoneal cysts were present in one patient each (Table 1). Thirty-six patients (9.2%) had multi-organ involvement and the mean number of cysts was 1.5 ± 1.0 . Fifty patients (12.7%) had spontaneous cyst rupture before the operation (Table 2).

Operative characteristics of the patients are shown in Table 3. Only 35 patients (8.9%) were ASA 3 (American Society of Anesthesiologists grade 3). Four patients refused surgery and 2 patients were referred to higher centers. Of the remaining 387 patients, 335 (85.2%) received general anesthesia and intubation, 9 patients (2.3%) received general anesthesia and Laryngeal Mask Airway (LMA), 39 patients (9.9%) received sedoanalgesia and 4 patients (1%) had regional anesthesia. In terms of types of treatment, 240 patients (59.6%) underwent laparotomy, 30 patients (7.4%) received laparoscopy and 73 patients (18.1%) underwent thoracotomy, while PAIR (percutaneous aspi-

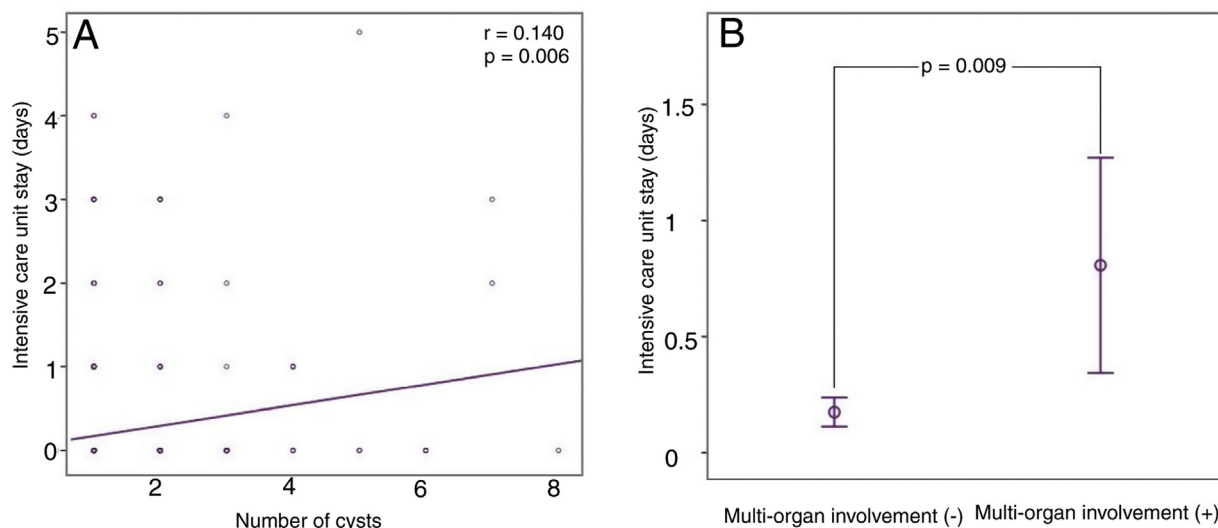


Figure 3 (A) Correlation analysis between number of cysts and intensive care unit stay. (B) Length of intensive care unit stay according to multi-organ involvement.

ration, injection and respiration) was performed in 60 patients (14.9%). Seventy-five patients (19.1%) had perioperative/postoperative need for blood products and 48 patients (12.2%) required intensive care unit (Table 4). The mean ICU stay was 1.9 ± 1.1 days in patients requiring ICU. The correlation analysis revealed that as the number of cysts increased, the length of ICU stay increased significantly ($r=0.140$, $p=0.006$) (Fig. 3A). In addition, the mean ICU stay was significantly higher in patients with multi-organ involvement than in patients without multi-organ involvement (0.8 ± 1.4 vs. 0.2 ± 0.6 days, $p=0.009$) (Fig. 3B).

During the perioperative period, 6 patients (1.5%) developed allergic reactions, 2 patients (0.5%) had diaphragmatic injury, 2 patients (0.5%) had cyst rupture, 1 patient (0.2%) had hepatic injury and 1 patient (0.2%) had hemorrhage due to vascular injury. Only 1 patient (0.2%) died during the operation. This patient had isolated cardiac hydatid cyst and developed anaphylactic shock due to cyst rupture during the operation. The most common complications in the early postoperative period were atelectasis (3.3%), biliary fistula (3.0%) and abscess (2.5%). The mean postoperative follow-up was 40 ± 17 months. Recurrence was observed in 33 patients (8.4%) during this follow-up period.

Discussion

Hydatid cyst has a worldwide distribution and is still a serious health problem in undeveloped and developing countries. Mediterranean, Middle East, Eastern Europe, Australia, South America, and African countries are endemic regions for hydatid disease. In Turkey, the disease is more frequently seen in the Eastern and Southeastern Anatolia regions where animal husbandry is common.²⁻⁵ Our study is important as it reflects the clinical outcomes of a region where hydatid disease is common and reports the anesthetic methods used during treat-

ment and the frequency of peri and postoperative complications.

Anesthesiologists play an important role in the evaluation of cyst hydatid cases due to determine the anesthetic methods used, management the developed complications and postoperative recover.⁹ All system evaluations must be performed exhaustively in preoperative period and high-risk patients such as requiring close follow-up should be determined. Anesthetic methods to be used may vary according to the localization of cysts, the clinic of the patient and the type of procedure. In this study, we performed general anesthesia, sedoanalgesia and regional anesthesia in the patients' management. Opioid analgesics, intravenous anesthetic drugs (propofol generally) and non-depolarizing muscle relaxant (rocuronium bromide) were used for general anesthesia, and anesthesia maintenance was continued with inhaled anesthetics (isoflurane or desflurane). In addition, opioid analgesics and intravenous anesthetic drugs were used for sedoanalgesia while bupivacaine was used for regional anesthesia.⁹ Pulmonary hydatid cyst cases were operated under lateral decubitus position by using double lumen endotracheal tube, central venous and arterial catheterization. To prevent allergy and anaphylactic reaction, dexamethasone and pheniramine were used prophylactically.

The disease is often asymptomatic since its progression is slow and the cysts are accidentally detected during routine evaluations. Patients usually present compression symptoms associated with cyst enlargement.⁶ The disease is usually diagnosed in the 3rd-4th decades of life. It is reported in the literature, however, that the disease may also be seen in children from 2 years-old onwards.¹⁰ Consistent with the literature, the majority of the patients were in the second and third decades of life and 72% of the patients were in the first 4 decades of life in our study. Additionally, the youngest patient was 3 years old and 10% of the cases were presented in the first decade of life.

After entering the body, the parasite enters the portal vein from the gastrointestinal tract and first reaches the

Table 3 Operative characteristics of the patients.

	N° of patients (n)	Percentage (%)
ASA score		
1	230	58.5
2	128	32.6
3	35	8.9
<i>Refusal of operation or referral to another center before the operation</i>	6	1.5
Anesthesia method		
General anesthesia + intubation	335	85.2
General anesthesia + LMA	9	2.3
Sedoanalgesia	39	9.9
Regional anesthesia	4	1.0
Treatment^a		
Surgery		
Laparotomy	240	59.6
Laparoscopy	30	7.4
Thoracotomy	73	18.1
Percutaneous PAIR	60	14.9
<i>Need for blood products</i>	75	19.1
Blood products used^b		
Erythrocyte suspension	145	
Fresh frozen plasma	49	
<i>Need for intensive care</i>	48	12.2

The values are expressed as number and percentage.

^a The same patient may have received more than one type of treatment.

^b The same patient may have received both erythrocyte suspension and fresh frozen plasma.

ASA, American Society of Anesthesiologists; LMA, Laryngeal mask airway; PAIR, percutaneous aspiration injection, reaspiration.

liver,¹¹ so the liver is the most commonly involved organ. The lung is the second most commonly affected organ after the liver. It is reported that in the lung, the right lobe is more commonly affected.¹² In our study, the majority of the patients (75.2%) had hepatic involvement. Pulmonary involvement was present in 19% of the patients and, particularly, the right lobe was more commonly involved in patients with pulmonary involvement. The spleen is the third most commonly involved organ after the liver and the lung, however, splenic involvement generally accompanies other organ involvements and isolated splenic involvement is more uncommon.¹³ In our study, there were 10 patients with splenic involvement but only 2 patients had isolated splenic involvement.

Cardiac hydatid cyst represents a very rare involvement and it is generally accompanied by other organ involvements.^{14,15} The most common cardiac location is the left ventricle due to its high vasculature. Right ventricular

Table 4 Complications observed in patients.^a

	N° of patients (n)	Percentage (%)
Peri-operative		
Allergic reaction	6	1.5
Diaphragmatic injury	2	0.5
Cyst rupture	2	0.5
Hepatic injury	1	0.2
Vascular injury	1	0.2
Death	1	0.2
Early postoperative period		
Atelectasis	13	3.3
Pneumothorax	5	1.3
Bronchospasm	3	0.7
Pleural effusion	1	0.2
Pneumonia	1	0.2
Abscess	10	2.5
Biliary fistula	12	3.0
Incisional hernia	4	1.0
Ileus	1	0.2
Late postoperative period		
Recurrence	33	8.4

The values are expressed as number and percentage.

^a Patients may have developed more than one complication.

and interventricular septal involvement are less frequent, while right atrial involvement is the most uncommon.¹⁶ The primary treatment is surgery, and early mortality following surgery (~10%) is quite high.^{14,17} Cardiac involvement was also rare in our study and only 2 patients (0.5%) had cardiac hydatid cysts. One of these patients was an 18 year-old male patient with both cardiac and hepatic involvement. This patient had interventricular septal involvement and he was transferred to another center at his own request due to high surgical risk. The other patient was also an 18 year-old male patient. This patient had isolated cardiac involvement and the cyst was located in the posterior wall of his left ventricle. This patient developed anaphylactic reaction due to cyst rupture during the operation. Bradycardia, hypotension, and hypoxemia occurred 1 mg adrenaline, 1 mg atropine, 8 mg dexamethasone, 45.5 mg pheniramine and a positive inotropic agent were administered, but the patient developed cardiovascular collapse. The patient did not response to the cardiopulmonary resuscitation and this patient was the only case of mortality in our series.

The presence of multiple cysts at the time of the diagnosis is one of the characteristic features of hydatid disease. There may be multiple cysts in the same organ and different organs may be affected at the same time. Studies showed that 10%–40% of the cases had multiple cysts.^{6,11} In our study, 111 patients (28.4%) had multiple cysts and 36 patients (9.2%) had multi-organ involvement. A 20 year-old male patient had the highest number of cysts. This patient had a total of 8 cysts in the liver and bilateral lungs. A 24 year-old female patient had the involvement of three organs, which was the highest number in our study. This

patient had a total of six cysts in the liver, spleen and bilateral lungs. In addition, we found in our study that ICU stay was significantly longer in patients with higher number of cysts and multi-organ involvement. Our results suggest that patients with multiple cysts may be high-risk patients requiring a close follow-up during the postoperative period. Therefore, in patients diagnosed with hydatid cyst disease, a detailed evaluation should be performed to detect multi-organ involvement and/or multiple cysts in the same organ. In patients with these conditions, the possibility of the need for postoperative intensive care should be kept in mind.

The treatment approaches for hydatid cyst disease are currently evolving with the advances in techniques, thus the anesthetic methods used in these cases are also changing. Surgery is the primary and the gold standard treatment and general anesthesia is generally used in these patients.^{2,11} Percutaneous treatment has been suggested as an alternative to surgery, and it can be as effective as surgical treatment in selected patients and is associated with high cure rates together with low complication, recurrence and mortality rates.^{18,19} Percutaneous treatment is mainly used in hepatic cyst hydatids and sedoanalgesia is more commonly preferred for anesthesia.³ In our study, 60 patients (14.9%) received PAIR and 57 of them had isolated hepatic cysts. Of the patients that underwent PAIR, 39 and 8 patients received sedoanalgesia and LMA, respectively, and an adequate level of anesthesia was achieved in these patients. All these results show that sedoanalgesia can be a reasonable anesthetic method in percutaneous procedures. We believe that PAIR can reduce hospital stay and costs in selected cases. Regarding the types of anesthesia used in our study, the great majority of the patients (85.2%), particularly those undergoing surgical treatment, received general anesthesia plus intubation.

Allergic reactions may be observed during the perioperative period due to the rupture of the cyst. The main cause of these reactions is that the highly antigenic content of cyst flows into the systemic circulation. Allergic symptoms vary from mild urticaria to anaphylactic shock, but anaphylactic shock is very rare.²⁰ In our study, allergic reactions were observed in 6 patients during the perioperative period; anaphylactic shock, in 1 patient; and urticaria in 5 patients. As mentioned previously, the patient who developed anaphylactic shock was administered with vasopressor, corticosteroid and antihistaminic, but did not respond to the cardiopulmonary resuscitation. Meanwhile, patients who developed urticaria were performed dexamethasone and pheniramine. After these treatments, all patients fully recovered from urticaria and no additional complication was developed.

In our study, the most common complications during the early postoperative period were atelectasis, biliary fistula and abscess. While the recurrence rates for hydatid disease vary between different series, some studies report a recurrence rate of 0.9%–22% in the 5 year follow-up.^{21,22} In our study, 8.4% of our patients experienced recurrence during the mean postoperative follow-up of 40±17 months. The recurrence rates in our study are similar to those in the literature. Although many mechanisms were suggested as reasons for recurrence, we believe that the most probable reason for recurrence in

our study was reinfestation due to social conditions of the region.

Summary

In conclusion, hydatid disease is still a global disease in undeveloped and developing countries. The anesthesiologist has a crucial role in the management of these patients. Before any intervention, patients diagnosed with hydatid cyst disease should always be evaluated in terms of multi-organ involvement and multiple cysts in the same organ, which can provide information about the need for postoperative ICU. The type of treatment procedure and localization of cysts determine the anesthetic management. Sedoanalgesia is more commonly used in percutaneous procedures, while surgical procedures are mostly performed under general anesthesia and intubation.

Conflicts of interest

The authors declare no conflicts of interest.

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