



Surgical Approach to Childhood Hydatid Cyst Disease: Experience of Two Centers

Çocukluk Çağı Kist Hidatik Hastalığına Cerrahi Yaklaşım: İki Merkez Deneyimi

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Abstract

Objective: We aimed to compare and evaluate our surgical approaches to childhood hydatid cyst (HC) disease by examining the diagnosis, follow-up and treatment records of patients with HC who underwent open surgery-percutaneous intervention.

Material and Methods: Demographic, clinical, laboratory, treatment and follow-up findings of 35 patients with HC requiring surgical intervention between May 2010 and May 2020 were analyzed retrospectively.

Results: Thirty-five patients were included in the study. Abdominal pain was seen at a rate of 77.1% in the children, chest pain at a rate of 13.2% and cough at a rate of 5.3%. One patient (2.8%) presented with dyspnea. There was only lung involvement in 21.1%, only liver involvement in 60.9%, liver and lung involvement in 7.9%. In one (2.6%) patient, only spleen and in one (2.6%) patient, liver and kidney involvement were detected. Thoracotomy was performed in patients with lung involvement. Laparotomy was performed in 12 (40%) patients with liver involvement, catheter in 10 (33.3%) and percutaneous aspiration-injection-re-aspiration (PAIR) in eight (26.6%) patients. Post-operative atelectasis developed in two (40%) patients after thoracotomy and one (8.3%) after laparotomy. Wound infection developed in three (25%) patients after laparotomy. When length of hospital stay after liver interventions was compared, there was a statistically significant shorter hospitalization time ($p < 0.001$) in the patients that underwent PAIR compared to the patients that underwent open surgery. However, there was no statistically significant difference between catheterization and open surgery ($p = 0.54$). Recurrence occurred in two patients with lung involvement and five (16.6%) patients with hepatic involvement. When the first attempts to the liver (laparotomy, catheter, PAIR) and re-

Öz

Giriş: Çalışmamızda, açık cerrahi-perkütan girişim uygulanan kist hidatik (KH) hastalarının tanı, takip ve tedavi kayıtlarını incelenerek, hastalara uygulanan cerrahi yaklaşımlarımızı karşılaştırılmalı olarak ortaya koymayı ve değerlendirmeyi amaçladık.

Gereç ve Yöntemler: Mayıs 2010-Mayıs 2020 yılları arasında cerrahi girişim gerektiren 35 KH'li hastanın demografik, klinik, laboratuvar, tedavi ve takip bulguları geriye dönük olarak incelendi.

Bulgular: Çalışmaya dahil edilen 35 hastanın 20 (%52.6)'si erkek, 15 (%42.9)'i kızdı. Yaş ortalaması 12 ± 3.13 (5-17) yıl olarak saptandı. Başvuru nedeni %77.1 ile karın ağrısı, %13.2 ile göğüs ağrısı, %5.3 ile öksürüktü. Bir olgu (%2.8) ise nefes darlığı ile başvurmuştu. Hastaların %21.1'inde sadece akciğer tutulumu, %60.6'sında sadece karaciğer tutulumu, %7.9'unda karaciğer ve akciğer tutulumu vardı. Bir (%2.6) hastada sadece dalak ve bir (%2.6) hastada karaciğer ve böbrek tutulumu birlikte saptandı. Akciğer tutulumu olan hastalara torakotomi; karaciğer tutulumu olan 12 (%40) hastaya laparotomi, 10 (%33.3) hastaya kateter ve sekiz (%26.6) hastaya perkütan aspirasyon injeksiyon reaspirasyon (PAIR) uygulandı. Torakotomi yapılan iki hastada (%40) ve laparotomi yapılan bir hastada (%8.3) post operatif atelektazi gelişti. Laparotomi yapılan üç (%25) hastada yara yeri enfeksiyonu gelişti. Karaciğere yapılan girişimler sonrası hastanede kalış süreleri kıyaslandığında PAİR uygulanan hastalarda açık cerrahi uygulanan hastalara göre istatistiksel olarak anlamlı kısa yatış süresi ($p < 0.001$) vardı. Kateter uygulanan hastalarla açık cerrahi girişim uygulanan hastalar arası istatistiksel anlamlı fark saptanmadı ($p = 0.54$). Akciğer tutulumu olan bir hastada (%20), karaciğer tutulumu olan beş (%16.6) hastada rekürrens gelişti. Karaciğere yapılan ilk girişimler (la-

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currence were compared, no statistical difference was found in any of the interventions (respectively, $p=0.63, 0.57, 0.37$).

Conclusion: Hydatid cyst affects many organs. It most commonly affects the lungs and liver. Although the number of our patients is limited, our study revealed that the use of percutaneous methods gives effective and reliable results in pediatric liver HC requiring surgical treatment.

Keywords: Hydatid cyst, child, organ involvement, surgery

Introduction

Hydatid cyst (HC) is a zoonotic disease that occurs with *Echinococcus* species (1). HC disease is endemic in Türkiye with an incidence of 3.4/100.000 (2). Age of the patient, structure of the cyst, location and size are important in treatment approach. The treatment can be medical (albendazole or mebendazole), surgical (open or laparoscopic), and with percutaneous interventions (2,3). Open surgical approach has been the gold standard method for a long time in large and complicated cysts (4,5). However, in recent years, percutaneous interventions have come to the fore as potential alternatives to surgery due to their efficacy, reliability, and low morbidity and mortality rates (5,6).

In this study, it was aimed to examine the diagnosis, follow-up and treatment records of HC patients having undergone open surgery and percutaneous intervention in our department and to comparatively present and evaluate our surgical approaches to childhood HC patients.

Materials and Methods

In our study, medical records of patients having undergone surgical intervention between May 1, 2010 and May 1, 2020 were retrospectively reviewed. Patients who only received medical treatment, cases who did not require surgery or those whose necessary parameters could not be reached (lost to follow-up, treatment continued in another center) were excluded from the study.

Hydatid cyst (HC) diagnosis was made by clinical, radiological and serologic tests. If deemed necessary, the patients, prior to treatment, had lung X-ray, abdominal or thoracic ultrasonography (US), thoracic or cranial tomography-magnetic resonance imaging and echocardiography. The patients were serologically evaluated with indirect hemagglutination test (IHA). Patients with a IHA value of $\geq 1/320$ were considered positive for *Echinococcus* infection. Lung HCs were classified according to the classification of World Health Organization's Informal Working Groups on Echinococcosis (WHO-IWGE) (7-9).

In our clinics, we apply medical treatment for cysts smaller than 5 cm and for cysts larger than 5 cm, if the location is suitable for surgery, we perform surgical treatment or medical treatment combined with percutaneous treatment.

parotomi, kateter, PAIR) ve rekürrens ilişkisi birbirleriyle kıyaslandığında hiçbir girişimde istatistiksel olarak fark saptanmadı (sırasıyla, $p=0.63, 0.57, 0.37$).

Sonuç: Kist hidatik birçok organı tutmakla birlikte en sık karaciğer ve akciğeri etkiler. Hasta sayımız sınırlı olmasına rağmen çalışmamız, cerrahi tedavi gerektiren çocukluk çağı karaciğer KH'lerinde perkütan yöntemlerin kullanımının etkin ve güvenilir sonuçlar gösterdiğini ortaya koydu.

Anahtar Kelimeler: Kist hidatik, çocuk, organ tutulumu, cerrahi

Open surgical intervention with posterolateral thoracotomy was performed in all lung HCs. During surgery, the surrounding tissues were protected with gauze bandage wetted with scolicidal agent [hypertonic (20%) saline solution]. Cyst content was aspirated and cystostomy was performed after scolicidal agent was applied. Germinative membrane was removed. During thoracotomy, tube thoracostomy was performed in all patients. Evaluation with a chest X-ray was made following the cessation of drainage from the chest tube, and the tube was decided to be removed or not.

In the treatment of liver HC, percutaneous aspiration injection reaspiration (PAIR), catheter or open surgery was used. During open surgery treatment, cyst content was aspirated and the scolicidal agent [hypertonic (20%) saline solution] was injected into the cyst. Germinative membrane was removed. The relation between cyst cavity and the biliary system was assessed. If the biliary tract was seen open, then the cavity was sutured and closed. In applicable patients, capitonnage or omentoplasty was performed on the remaining cavity. In a patient with spleen cyst, the surgical procedure that we implement on liver HC was performed. Omentoplasty was performed on the remaining cavity. Capitonnage could not be performed in patients with infected cysts. A drainage catheter was placed in these patients. Same procedure with laparotomy was applied to the liver cyst of a patient with liver and kidney involvement. Cystostomy was performed on the kidney cyst so as not to harm the surrounding cavity walls and neighboring large vessels. Total excision was carried out without capitonnage.

PAIR-catheter procedures were done with US. During PAIR, cyst content was aspirated with the help of a needle, the scolicidal agent [hypertonic (20%) saline solution] was injected, and after 10 minutes, reaspiration was done. PAIR was conducted in cysts smaller than 6 cm or with a fluid content under 100 cc. Catheter treatment was applied to cases with cysts larger than 6 cm and with a cyst content over 100 cc. During catheter procedure, drainage catheter, 8 G in size, was inserted into the cyst content and was kept until catheter drainage was below 10 cc in 24 hours. Afterwards, cavitography was taken, sclerosing agent was injected, and the catheter removed.

In two cases with lung and liver involvement, the lung cyst was treated first with thoracotomy, and one month later, PAIR was implemented on the liver cyst.

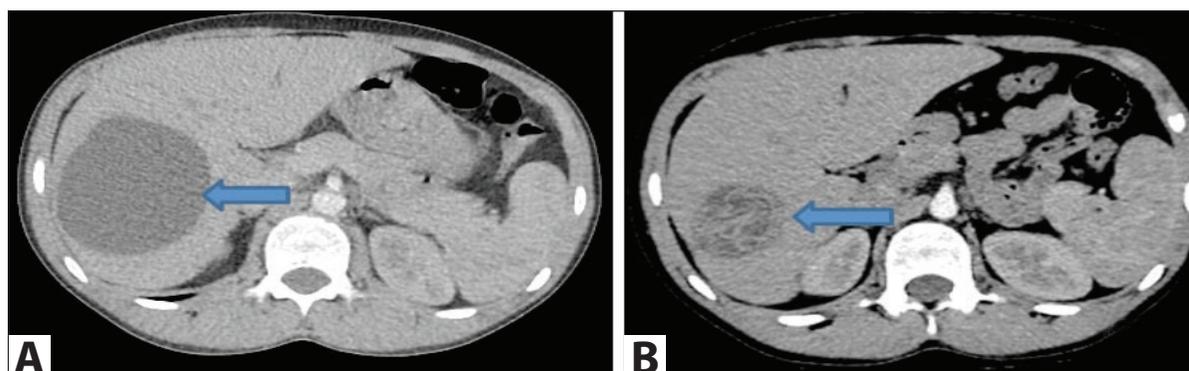


Figure 1. Abdominal tomography image of a patient undergoing PAIR.

A. WHO type 1 hydatid cyst on the 6th-7th segment of the right lobe of the liver. **B.** Tomography image in the 9th month after PAIR.

Albendazole (10 mg/kg/day) treatment was started on patients planned to undergo surgery three weeks before the operation. Treatment response was evaluated by clinical, radiological and serologic tests. Patients who had been discharged were evaluated with liver function tests once in three weeks, and albendazole treatment was completed in three months. The patients were called to follow-up in postoperative 1st, 3rd, 6th, 9th and 12th months and every six months afterwards. With the amelioration of clinical findings, the decrease in the diameter of the cyst, collapse of the cyst, progressive increase in the echogenity of the cyst fluid and disunion of the membranes from the capsule were considered treatment success (9). New cyst formation, recurrence of the cyst, increase in the dimension of the cyst or remaining unchanged were considered as treatment failure. Figure 1 demonstrates the tomography image of a case in whom PAIR was performed in the postoperative 9th month.

The study was carried out in accordance with the 2008 Helsinki Declaration and having received ethics approval from both centers (Date: 27/10/2021, Number: 2021/217). Informed consent was not obtained due to the retrospective nature of the study.

Statistical analyses were performed on IBM SPSS (Windows Version 21.0. Armonk, NY: IBM Corp.). Continuous variables were expressed with median (range) and categorical variables with frequency (percentage). Mann-Whitney U test was used in the analysis of continuous variables since parametric assumptions were not met. Chi-square test was used in the analysis of nominal variables. In the analysis of nominal values, Fisher's Exact test was used when the distribution was not compatible with Chi-square test. $p < 0.05$ value was accepted statistically significant.

Results

There were 35 patients, with 20 girls (52.6%) and 15 boys (42.9%). Mean age was 12 ± 3.13 (5-17) years. The most common presentation reasons were abdominal pain with 77.1%,

Table 1. Cyst localizations of the patients

Localization	Patient number (n= 35, %)
Liver right lobe	22 (55.3%)
Right lung mid-lower lobe	3 (7.9%)
Lung and liver	3 (7.9%)
(Liver left lobe-lung left lower lobe; liver right lobe-left lung lower lobe; liver left lobe-lung left upper lobe)	
Left lung lower lobe	2 (5.3%)
Liver left lobe	2 (5.3%)
Left lung upper lobe	1 (2.6%)
Spleen	1 (2.6%)
Liver and kidney	1 (2.6%)

chest pain with 13.2%, and cough with 5.3%. One case presented with shortness of breath.

Of the cases, 21.1% had only lung involvement, 60.6% had only liver involvement, and 7.9% had lung and liver involvement. Spleen involvement was detected in one patient (2.6%), and liver and kidney involvements were found in one patient (2.6%). Table 1 summarizes cyst localizations of the cases.

On presentation, echinococcus IHA test was positive in 21 of the patients (60%) and negative in 14 patients (40%). Mean cyst size was found as 73.2 ± 21.7 (51-126) mm.

Thoracotomy was performed in patients with lung involvement, laparotomy was performed in 12 patients (40%) with liver involvement, catheter and PAIR were performed in 10 (33.3%) and 8 (26.6%) patients, respectively. Preoperative cyst rupture developed in 4 patients (13.3%) with liver involvement. Upon seeing bile juice from the drain postoperatively in a patient having undergone laparotomy, follow-up with the drain was extended. On postoperative 7th day, upon finding nothing in the drain, the drain was removed being evaluated by US. The patient was then discharged. Postoperative atelectasis developed in two thoracotomy patients (40%) and in one

laparotomy patient (8.3%). Wound site infection developed in three laparotomy patients. Length of hospital stay for open surgery patients was mean 5.5 ± 1 (5-7) days for lung HC and 4.5 ± 1.6 (3-8) days for liver HC. Length of hospital stay in patients undergoing PAIR and catheter were 2.2 ± 0.4 (2-3) days and 3.7 ± 0.9 (3-5) days, respectively. When length of hospital stay after operation to the liver was compared, a statistically significantly shorter length of stay was detected in PAIR patients compared to open surgery patients ($p < 0.001$). A statistically significant difference was not observed between cases undergoing catheter and open surgery ($p = 0.54$).

Due to recurrence in a patient with lung involvement (20%), second thoracotomy was performed. Recurrence was seen in 5 patients (16.6%) following first intervention in patients with liver involvement. When the relation between first interventions (laparotomy, catheter, PAIR) and recurrence was compared to one another, a statistical difference was not detected (respectively, $p = 0.63, 0.57, 0.37$). In two patients with recurrence, catheter was implemented as first intervention and then laparotomy was done. In a PAIR patient, cure was obtained after a second PAIR, and a patient who developed recurrence following laparotomy was cured with catheter application. Mean follow-up period of the patients was 54.1 ± 35.5 (3-120) months.

Discussion

Hydatid cyst may involve many organs (3). Organ involvement may show difference according to regions and age (4). In publications from various countries, while there are studies reporting lung involvement the most in terms of localization in the childhood, there are also studies presenting otherwise and reporting liver involvement to be the most common (3,10). We are of the opinion that the differences in the organs involved in studies related to HC originate from epidemiological factors. In our study, the most commonly involved was the liver in cases requiring surgery.

Symptoms vary according to the organs involved (3). Symptoms such as chest pain, cough and shortness of breath are seen in lung involvement, whereas, in liver involvement, symptoms such as fullness, abdominal pain and vomiting are encountered (11). However, HC may have an asymptomatic course in children (12,13). CH is accidentally detected in these patients (3,12,13). In our girl patient with spleen involvement, HC was accidentally detected on abdominal US taken after abdominal trauma. In a case with liver and kidney involvement, the patient had no symptoms regarding the kidney. However, most of our patients showed symptoms according to the organ involved. The most common symptoms in patients with liver and lung involvement were abdominal pain and chest pain, respectively.

The most frequently used agents in medical treatment are albendazol and mebendazole (4). However, surgery and

percutaneous interventions are at the forefront in large and complicated cysts. Initiating medical treatment prior to open surgery and percutaneous interventions is controversial (3). However, studies have reported that medical treatment prior to and after open surgery and percutaneous interventions increases efficacy of the treatment (14,15). Although there is no consensus on how long medical treatment should be continued after surgery, there are studies reporting that treatment can be prolonged for a year (14). In our study, patients were started on medical treatment three weeks prior to surgery, and the medical treatment lasted at least three months post-operatively.

In selected cases, percutaneous methods have started to be used as alternatives to open surgery (5,6). Percutaneous treatment is not recommended in lung HC since there is not sufficient evidence of efficacy and reliability (3,16). Therefore, we performed open surgery on all lung HC patients. According to WHO-IWGE, percutaneous treatment should be used in liver HC of CE1 and CE3A types (17). Percutaneous treatment can be performed by catheterization and PAIR. Turkish Radiology Society recommends PAIR in cases under 6 cm and catheter in cases over 6 cm (18). In our study, catheter was applied to patients with a cyst over 6 cm, fit for percutaneous treatment. When patients undergoing PAIR were compared to open surgery patients, length of hospital stay was detected to be distinctively shorter. A statistical difference was not found in terms of length of hospital stay in catheter treatment. When complications of the interventions were assessed, wound site infection and atelectasis were not found in patients undergoing percutaneous treatment.

Recurrence rates in HC were 0-25% (19). Recurrence rate in our study was 14.3%, and recurrences developed after thoracotomy in one patient, after PAIR in other 4 patients, after laparotomy in one and after catheter treatment in two patients. When interventions to the liver were compared, significant difference was not detected in recurrence rates. These results make us consider that percutaneous interventions are as efficient as open surgery in terms of liver HC.

Conclusion

Although hydatid cyst involves many organs, the lung and liver are the two most commonly involved ones. Despite limited number of patients in our study, we put forth that percutaneous methods show efficient and reliable outcomes in childhood liver HCs requiring surgical treatment.

Ethics Committee Approval: This study was approved by İstanbul University of Health Sciences Sancaktepe Şehit Prof. Dr. İlhan Varank Training and Research Hospital Scientific Research Ethics Committee (Decision no: 2021/217, Date: 27.10.2021).

Informed Consent: Patient consent was obtained.

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