Surgery for Cardiac Hydatid Disease: an Anatolian Experience

Kardiyak Hidatik Hastaların Cerrahisi: Anadolu Deneyimi

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Abstract

Objective: The purpose of this study was to describe the clinical/pathological features and the outcome of the surgical treatment of cardiac hydatid disease in our unit and other hospitals of Anatolia over a fifteen-years period.
Methods: Between 1984 and 2001, fifty cases of surgically treated cardiac hydatid disease were identified by systematic literature search from Anatolia. Twelve patients were operated at the Department of Cardiovascular Surgery, Ankara University within the same period. Overall thirty-nine patients were female (mean age 29.8 ±14 years). Sixty-three percent of patients were complaining of dyspnea at the time of referral to the hospital and 22% presented with signs of acute coronary syndrome.
Results: The most common cardiac location was the left ventricle (46.7%) followed by the interventricular septum, the right ventricle and atria. The most common procedure was controlled puncture and aspiration of the cyst content, its excision and closure of the resulting cavity, which were performed under cardiopulmonary bypass. Overall Anatolian operative mortality was 4.8% (3 patients). No late deaths but recurrence in one patient have occurred.
Conclusion: In view of the lack of efficient alternative treatment options, we recommend surgical intervention even in asymptomatic patients in order to prevent the occurrence of lethal complications. *(Anadolu Kardiyol Derg 2003; 3: 238-44)*
Key Words: Cardiac hydatid disease, echinococcus, cystectomy, albendazole

Özet

Amaç: Anadolu hastanelerinde ve kendi ünitemizde, on beş yıllık bir dönemde yayınlanmış kardiyak hidatik hastalığın klinik ve patolojik değerlendirmesinin yapılması, cerrahi tedavi sonrası sonuçlarının ortaya konması.
Yöntemler: Bin dokuz yüz seksen dört ile 2001 tarihlerini kapsayan sistematik literatür araştırması sonrasında cerrahi tedavi görmüş 50 kardiyak hidatik hastanın varlığı saptanmıştır. Aynı dönemde Ankara Üniversitesi Tıp Fakültesi, Kalp ve Damar Cerrahisi Anabilim Dalında, 12 kardiyak hidatik vakası ameliyat edilmiştir. Tüm hastalar birlikte değerlendirildiğinde olguların otuz dokuzunu kadınlar oluşturmaktaydı (ortalama yaş 29.8 ±14 yıl). Olguların %63'ü dispne, %22'si akut koroner sendrom bulguları ile başvurmuşlardı.
Bulgular: Kardiyak hidatik hastalığın en sık gözlendiği kalp boşluğu sol ventrikül iken (46.7%), bunu sırasıyla interventriküler septum, sağ ventrikül ve atriyumlar izlemiştir. En sık uygulanan cerrahi tedavi kardiyopulmoner baypas kullanılarak yapılan kist içeriğinin kontrollü aspirasyonu, eksizyon ve geriye kalan kavitenin kapatılması olarak belirlenmiştir. Araştırmaya dahil edilen tüm Anadolu kist hidatik hastalarında operatif mortalite %4.8 olarak belirlenmiştir (3 hasta). Uzun süreli takiplerde hidatik hastalığa bağlı ölüm bildirilmezken, bir olguda nüks ortaya konmuştur.
Sonuç: Diğer alternatif tedavi seçeneklerinin yetersizliği nedeniyle, asemptomatik kardiyak hidatik olgularında bile ölümcül komplikasyonlar ortaya çıkmadan, erken cerrahi tedavi uygulanmasının gerekliliğini vurguluyoruz.

Introduction

Echinococciasis is a tissue infection of humans caused by the larval stage of Echinococcus granulosus, E. multilocularis, E. oligarthrus, or E. vogel. The incidence of hydatid disease is 1:2000 in Turkey. Cardiac involvement is rare, occurring in about 0.5 to 2% of cases (1). Since the introduction of cardiopulmonary bypass, several successful surgical cases were reported worldwide. Unless the disease is recurrent or inoperable, patients with cardiac hydatid disease must undergo surgery to avoid life-threatening complications such as cyst rupture, anaphylactic shock, tamponade (2), pulmonary (3, 4), intracereb-

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ral (5) or peripheral arterial embolism (6), acute coronary syndrome (7, 8), arrhythmias (9) and infection (10). The efficacy of alternative medical therapies is not well established.

The aim of this work was twofold; 1) to study the clinical/pathological features and the indications and results of surgery over 15 years in our unit and 2) to collectively analyse our patients with the published cases from other hospitals in Turkey over the same time period.

Materials and Methods

Between November 1984 and December 2001, a total of 12 patients with cardiac hydatid disease underwent sternotomy at the Department of Cardiovascular Surgery of Ankara University Medical School (AUTF), Turkey. There were four male and eight female patients, with a mean age of 31 ± 12 years (range 4 to 52 years). Medline search within the same period identified 50 patients from Anatolia undergoing surgical treatment for cardiac hydatid disease (Table 1). Thus, a total of 62 patients undergoing surgery for cardiac hydatid disease are the subjects of this report. Three cases reported as having medical treatment within the same period were not included in this study. The patients were analysed with regard to the demographic, clinical presentation, type of surgical resection, operative mortality, recurrence, late complications and long-term survival. Patients' data are shown in Table 1. In the combined analysis, there were 39 female and 23 male with age ranging from 4 to 70 years. Patients` charts from our department and published reports were collectively reviewed.

Table 1. Reports on surgically treated cardiac hydatid disease in the Anatolian literature.

First Author / Year / Location	Period	No. of patients	Age	Gender (F/M)	Mortality
Birincioglu (21), Ankara	2001	1	9	0/1	0
Aydogdu (22), Antalya	2001	1	13	1/0	0
Telli (8), Konya	2001	1	40	0/1	0
Ceviz (10), Erzurum	2001	1	24	1/0	0
Ozer (6), Ankara	1985-1997	5	39 ± 24	3/2	0
Keles (23), Istanbul	2000	1	37	0/1	0
Karadede (24), Diyarbakır	2000	1	55	0/1	0
Kanadasi (25), Adana	2000	1	25	1/0	0
Kaplan (12), Istanbul	1988-1999	8	33±14	7/1	1/8
Birincioglu (2), Ankara	1977-1998	14	28±12	8/6	1/12
Salih (11), Adana	1988-1998	3	27±13	2/1	0
Erenturk (26), Istanbul	1998	1	34	1/0	0
Ege (4), Malatya	1997	1	41	1/0	1/1
Turgut (5), Aydın	1997	1	7	1/0	0
Alehan (27), Ankara	1995	1	11	0/1	0
Kulan (7), Trabzon	1995	1	13	1/0	0
Emirogullari (28), Kayseri	1995	1	12	0/1	0
Unal (29), Trabzon	1995	1	14	1/0	0
Pasaoglu (13), Ankara	1994	1	41	0/1	0
Akcakaya (30), Istanbul	1994	1	12	0/1	0
Yekeler (31), Erzurum	1993	1	23	0/1	0
Pasaoglu (32), Ankara	1992	1	27	1/0	0
Bayezid, (3), Istanbul	1991	1	32	1/0	0
Erol (14) Ankara	1984	1	23	1/0	1
Current series, Ankara	**1984-2001**	**12**	**29.8 ± 14**	**8/4**	**0**

Clinical Features and Preoperative Evaluation

Sixty-one percent of patients were complaining of dyspnea at the time of referral to the hospital. Other presenting symptoms of cardiac hydatid disease are shown in Table 2. Sinus rhythm was present in 52 (83%), atrial fibrillation in 6 (9.7%), right bundle branch block in 5 (8%) patients; two patients (3.2%) experienced ventricular arrhythmias and 1 patient (1.6%) had a permanent pacemaker. Preoperative evaluation in our series was done by means of physical examination, hematological and biochemical investigations, chest X-ray, electrocardiogram, computed tomography (CT) of the chest, echocardiography, magnetic resonans imaging (MRI) and coronary angiography. Additional investigations such as liver ultrasound, head or abdominal CT were performed as indicated by the clinical findings and/or laboratory parameters.

Casoni's intradermal test was carried out in 32 patients, a complement fixation test (CFT) in 24, an indirect hemagglutination test (IHA) in 18, enzyme-linked immunosorbent assay (ELISA) in 14 and an eosinophil count in 37 patients. Other diagnostic tests used were computed tomography in 17 patients (27.4%), magnetic resonance imaging in 12 (19.4%), coronary angiography in 6 (9.6%), pulmonary artery digital substraction angiography in 2 (3.2%) and myocardial scintigraphy in one patient (1.6%).

The heart was the only location of the disease in 38 patients (61.2%), whereas 12 patients (19%) had associated lung, liver, brain, and peritoneal, and/or renal hydatid cysts.

Table 2. Clinical manifestations of cardiac hydatidosis in cases from Anatolia

	No. of patients (*)	%
Dyspnoea	39 (7)	62.9
Palpitation	25 (5)	40.3
Chest Pain	14 (2)	22.5
Syncope	3 (2)	4.8
Cough	8 (2)	12.9
Hepatomegaly	7 (3)	11.3
Haemotysis	6 (2)	9.7
Pulmonary embolism	4 (1)	6.5
Acute abdomen	2 (0)	3.2
Peripheral embolism	3 (0)	4.8
Asymptomatic	3 (1)	4.8
Cyanosis	2 (1)	3.2
Cerebral embolism	2 (0)	3.2
Constrictive Pericarditis	1 (0)	1.6
Anaphylactic reaction	1 (0)	1.6
Cardiac tumor	1 (0)	1.6

(*): Patients from Ankara University School of Medicine

The Operation

At operation the aim was to achieve complete clearance of hydatid cyst without uncontrolled rupture. At exploration, the pericardium was carefully packed with pads around the cysts to reduce the risk of pericardial soil (Fig. 1). The operation was performed with median sternotomy in all patients except two cases who underwent posterolateral thoracotomy. Standard cardiopulmonary bypass (CPB) techniques using moderate hypothermia and cardioplegic arrest were used in 55 patients. Seven patients with subepicardial cysts within the ventricles were operated on without using CPB (2, 11). The cysts were reached via ventriculotomy in 51, atriotomy in 10 and trans-

Table 3. Location of cardiac hydatid disease.

Location	No. of patients (*)	%
Left ventricle	29 (5)	46.7
Interventricular septum	12 (1)	19.3
Right ventricle	12 (4)	19.3
Right atrium	7 (1)	11.2
Left atrium	1 (1)	1.6
Sinus of Valsalva	1 (0)	1.6

(*): Patients from Ankara University School of Medicine

Figure 1. Intraoperative photograph showing cardiac hydatid cyst located at the interventricular septum. The pericardium was packed with pads around the cyst to reduce the risk of pericardial soil. Following hypertonic saline application, controlled opening of the cyst was achieved.

aortic approach in one patient. There was no uniformity in the intraoperative use of scolicidal agents. Hypertonic saline (10% NaCl), chlorhexidine, 80% alcohol, 5% silver nitrate solution, 1% iodine solution were used as scolicidal agents.

Results

Preoperative evaluation

The results of serologic tests were variable and rather inconclusive. In 11 of the 32 patients (34.3%) Casoni's intradermal test was positive. False negative results were demonstrated in 10 patients (41.6%) for CFT, in 8 patients (44.4%) for IHA and in 3 patients (21%) for enzyme-linked immunosorbent assay (ELISA). Confirmation of diagnosis was obtained by echocardiography in 58 patients (94%) and computed tomography in 35 patients (56%).

Types of operations

All patients except two, who had posterolateral thoracotomy, underwent median sternotomy. Mean CPB time was 53 ± 18 minutes ranging between 28 and 83 minutes. Cardiopulmonary bypass with femoral cannulation and total circulatory arrest was used in one patient, with right ventricular hydatid cyst (12). Subendocardial cysts were more common in the right ventricle (9. 75%) and subepicardial cysts were predominant in the left ventricle (22.78%).

The most common procedure was cystectomy and capitonage or cystectomy alone. Associated procedures included: left ventricular patch in two patients (2, 12), patch repair of interventricular septum (AUTF), mitral valve replacement in two patients (5, 13), pulmonary embolectomy in two patients (4, 6), and femoral embolectomy in three patients (6).

The size of the cardiac hydatid cysts were measured and reported in 26 patients. Average cyst diameter was 5.4 ± 2.2 cm whereas average cyst volume was 112 mm3 (range of 4.86-480 mm3).

Operative mortality and morbidity

Overall operative mortality was 4.8% (3 patients). Causes of death were pulmonary embolism in two patients (12, 14), and rupture of interventricular septum in one patient (2). Major postoperative complications were requirement for permanent pacemaker implantation in two patients and re-opening for bleeding in one patient.

Recurrence and late survival

The average duration of follow-up at AUTF was 8.5 ± 5 years. During this follow-up period all patients had echocardiographic examination annually within the first 5 years after surgery. We did not detect any recurrence in AUTF patients however there was one recurrence reported by Kurtoğlu et al. (15) within 3 months of surgery despite medical therapy with Albendazole. Bayezid et al. (3) reported a patient who was admitted to hospital with congestive cardiac failure. This patient required further surgery for liver hydatid cysts 12 months after the initial operation. All of our patients are alive and there have been no late deaths in the reports from the other Anatolian institutions.

Discussion

The tapeworms, or cestods, are ribbon-shaped segmented hermaphroditic worms which inhabit the intestinal tract of many vertebrates. The term, hydatis, is the Greek word for a drop of water, which refers to the fluid-filled cysts formed by the Echinococcus species larvae in humans. Hydatid disease is endemic in most sheep-raising countries in Asia, Europe, South America, New Zealand, and Australia (16). The main form is due to Echinococcus granulosus. Most cases in Europe and North America occur in immigrants from highly endemic countries.

Like other cestods, echinococcal species have both intermediate and definitive hosts. The definitive host is a carnivore mainly dogs that harbours the adult tapeworm in the small intestine; the carnivore becomes infected by ingesting the larval form in tissue of the intermediate host. The intermediate hosts, chiefly herbivorous mammals and also humans, become infected by ingesting tapeworm eggs passed from carnivore faeces. The larval stage is referred to as a hydatid cyst. Human consumpti-

Figure 2. Intraoperative photograph demonstrating daughter vesicles following cystectomy.

on is mostly inadvertent via "hand-to-mouth" transmission occurring after close contact, such as petting, with infected animals. The ingested parasitic larvae migrate through intestinal mucosa and are carried to the liver by the portal venous circulation and lymphatics where most of the larvae are filtered out. They migrate to the host's viscera where they develop into mature larval cysts (Fig. 2). If embryos bypass the liver, they reach the lungs and other organs via systemic circulation or lymphatics. It is well documented that the majority (52%–77%) of hydatid cysts are located in the liver followed by lungs (9%–44%), the spleen (2-3%) (17), kidney (1-2.5%) (17), brain, and heart (0.5-2%) (18). Larvae reach the left side of the heart from the coronary circulation, patent foramen ovale, the lymphatics, or through the pulmonary veins. The host's dense fibrous response to the presence of parasite creates an adventitial pericyst layer.

Cardiac hydatid disease is seen in any age and sex group (1, 2,12), although it is more common in those 20 to 40 years of age. In our study the female/male ratio was 1.7/1 and the mean age of the patients was 30 years. The clinical presentation varies, depending on the location, size and integrity of the cardiac cysts. In previous studies, the most common locations of cardiac echinococci cyst were the left ventricle (60%), and the ventricular septum (9% to 20%), but the right ventricle and right atrium can also be involved (4% to 17%) (19). In agreement with previous reports, in our retrospective analysis from Anatolia, the left ventricle was the most common location (46.7%), followed by the interventricular septum (19.3%), right ventricle (21%), right atrium (9.7%), left atrium (1.6%) and sinus of Valsalva (1.6%). It is remarkable that isolated cardiac involvement was seen in 61% of the patients. The explanation of primary cardiac involvement can only be speculative and requires further research. In three quarters of all cases, the hydatid cyst is enlarged subendocardially in the right heart, and subepicardially in the left heart as confirmed in this meta-analysis.

There is no uniform clinical presentation of cardiac hydatid cysts; as it is shown in our study, patients may present with symptoms due to mechanical interference with cardiac function, simulating coronary artery disease, arrhythmias, conduction disturbances, pericarditis, and peripheral emboli or as an abnormality of the cardiac silhouette on chest x-ray. The most frequent symptom was dyspnoea followed by palpitations, angina, syncopal episodes according to the data collected.

Because cardiac hydatid cysts can cause life-threatening complications such as cardiac failure, cyst rupture, embolization etc, the establishment of an early diagnosis and the performance of a timely, potentially curative, surgical intervention are of paramount importance. The differential diagnosis of an intracardiac cyst should include cardiac echinococcal (hydatid) cyst, in patients from sheep-raising countries where Echinococcus infestation is endemic. Given the ease of global mobilization, physicians from Western countries also need to consider this worldwide problem in differential diagnosis. Serologic examinations have low diagnostic sensitivity and specificity and have only limited use. Six of the12 cases from AUTF demonstrated negative serology. In agreement with previous reports, this series show that a negative serology cannot rule out the diagnosis. Eosinophilia is uncommon except after cyst rupture. In this study, echocardiogram provided definitive diagnosis in 94% of the cases whereas angiogram was an essential diagnostic modality in patients with symptoms of acute coronary syndrome. Two patients required coronary angiography for angina-like symptoms but demonstrated normal coronary anatomy at the AUTF. Seven patients were evaluated with transoesophageal echocardiography, but in one case, the echoluscent and multiseptate nature of hydatid cyst was absent and the patient was further evaluated using MRI. The latter provided not only anatomic extent and position of the mass and its relation to cardiac chambers but also multiorgan involvement. A low-intensity rim with a thickness of 4 mm was present in this case, which was helpful to reach correct diagnosis.

Information in the literature on medical treatment of cardiac hydatid disease is limited. Benzimidazole carbamate is a viable option for symptomatic cysts that are inoperable. Mebendazole was one of the first in this class to show efficacy (1971), but it has been quickly replaced by albendazole, which is better absorbed. Response to this therapy is apparently related to the thickness of the cyst wall, which drug must penetrate to reach the germinal layer. Therapy is usually in cycles of 28 days on treatment and 14 days off with a dosage of 10–15

mg/kg or 400 mg twice a day. In recent years albendazole have been used in Turkey commonly for 4 to 8 weeks before and after surgery to reduce the risk of metastatic spread during the operation. However the side effects from the drugs are considerable including the rupture of the hydatid cyst or abscess formation. Common side effects of Albendazole are abdominal pain, diarrhea, elevated liver enzymes and allergic reactions. Eight patients from AUTF received Albendazole after the diagnosis and postoperatively since the drug became available in Turkey (1994) whereas other four patients had been operated before this period.

Standard median sternotomy and cardiopulmonary bypass are well established modalities for surgical treatment of cardiac hydatid disease. However subepicardial cysts can be operated with off-pump technique. Although there was no mortality among AUTF patients, Anatolian literature review revealed 3 deaths due to pulmonary embolism (2), and rupture of interventricular septum respectively. Overall postoperative mortality rate of 4.8% is comparable with mortality figures quoted from elsewhere (1, 20). Postoperative progress is usually satisfactory and uncomplicated but involvement of the interventricular septum may result in complete heart block and need for a permanent pacemaker as it was the case in two of our patients. Non-surgical conservative approach has been recommended for only asymptomatic high risk patients with small and completely calcified cysts if no adverse effects on the hemodynamics or blood supply to the heart could be proven (16).

The study suffers from the weakness inherent to any retrospective study, including potential inconsistency of data captured over time and acquisition of postoperative late events. Secondly published hydatid cyst cases may not reflect the characteristics of all Anatolian hydatid cyst population. We believe that the case reports mentioned in this analysis have been chosen for their originality by their authors and may not reflect the institutions' all series. However case reports provided sufficient morphological features, clinical and anatomical description. Despite the limitations of this study, we feel that the data presented here are sufficient to support the importance of an early diagnosis and early surgical treatment of cardiac hydatid disease.

Conclusions

This study suggests that surgical resection of cardiac hydatic cysts offers a good chance of cure with acceptable operative mortality. Cystectomy alone or with capitonage appears to be effective in preventing recurrence in the absence of multiorgan involvement. In view of the lack of efficient alternative treatment options, we recommend surgical intervention even in asymptomatic patients in order to prevent the occurrence of lethal complications. Serial echocardiographic examinations or other imaging modalities in the follow-up should be considered to detect recurrences.

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