

Review Article

Cardiac Hydatid Cysts- Review of Recent Literature

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- Albendazole

Abstract

Echinococcus is a parasitic source of infection that occurs in humans in endemic regions of the world. It causes medical, veterinary and economic problems for endemic developing countries. Primary echinococcosis of the heart represents 0,5-2% of all hydatid disease. This is a retrospective review of recent literature that consists information about 35 case report studies from different regions and aims to emphasize the significant points of approach principals to cardiac echinococcosis.

ABBREVIATIONS

CT: Computerized Tomography; MRI: Magnetic Resonance Imaging; IHA: Indirect Hemagglutinin; ELISA: Enzyme-Linked Immunosorbent Assay

INTRODUCTION

Human cardiac echinococcosis is of worldwide severity and causes medical, veterinary and economic problems for endemic developing countries. The diagnosis of cardiac echinococcosis can be hard and must be suspected in any patient from endemic or farming areas. It should be considered in the differential diagnosis of tumoral and cystic masses and treat properly with combination of different methods. We report a review of recent literature, and we aim to emphasize the significant points of approach principals to cardiac echinococcosis.

DISCUSSION

Review

We retrospectively reviewed the latest reports of cardiac echinococcosis cases from different regions of the World between 2014 and 2017. Searching the terms: 'cardiac hydatid cysts and cardiac echinococcosis'. All papers in English were included. 41 were females and 45 were males. 42 patients lived in Turkey and, the others were from Iran (five cases), Saudi Arabia (one case), Italy (two cases), Pakistan (one case), China (26 cases), South West Bengal (one case), Greece (one case), Tunisian (one case), India (four cases), Peruvian (one case) [1-35]. Cases are described in detail on (Table 1).

Five patients had refused to undergo surgery, they had been treated with chemotherapy of albendazole. Others were operated for total excision of cysts in elective surgery under cardiopulmonary bypass.

Albendazole was used in 77 patients pre and postoperatively,

mebendazole was used in one patient. We could not have data from reports about medication of eight patients.

At postoperative course, all survived and operated cases had good health, and had no recurrence. Two patients had died, one of them was because of anaphylactic shock during surgery and one was immediate death while on the waiting list for surgery.

Etiology and epidemiology

Echinococcus is a parasitic disease that occurs in humans in endemic regions of the world and is caused by the larval stage of Echinococcus granulosus, E. multilocularis, or E. vogeli [35]. Infection arises from handling dogs or ingesting cyst-containing meat from an intermediate host in an endemic area. Echinococcus multilocularis is adjusted to circulate between wild and domestic canids as definitive hosts and small mammals as intermediate hosts. Small mammals combine limited space for the metacystode growth with a short life time, so that morphological differences to metacystodes of other Echinococcus spp. (compact, vesicular growth with high density of protoscoleces) and the short time needed for the development of protoscoleces can be explained as an evolutionary response to conditions. In the largest part of the endemic area E. multilocularis life cycles are based on rodents, predominantly voles (Arvicolinae), and different species of canids that prey on them. Based on field data and/or experimental infections, other families of Carnivora are caused to be partially or completely refractory to infection. The high number of human cases certainly reflects the wide distribution and high frequency in dogs and livestock, but an apparently low specificity at the intermediate host level may also contribute to an enhanced infectivity or pathogenicity for humans compared to other Echinococcus spp. causing cystic echinococcosis. Epidemiological data suggest that this species is particularly well adapted to sheep as intermediate hosts, which is reflected in high prevalence [36]. That seems as one of the main reasons of the prevalence and severity of Echinococcus manifestations.

Table 1: Studies of cardiac echinococcosis world wide.

Author	Age Gender Region	Symptoms	Size of cyst	Localisation	Serology	Surgery	Medication	Follow up
Poorzand H. et al. 2014 (1)	29 Female Iran	Shortness of breathe	50x47 mm	RV	+	OHS	Albendazole 400 mg/day	Good, 1 year
Gocen U. et al. 2014 (2)	6 Female Turkey	Chest pain Dyspnea	40x30 mm	IVS	+	OHS	Albendazole 400 mg/day	Good, 6 mo.
Alshehri H. et al. 2014 (3)	29 Female Saudi Arabia	Palpitation	70x60 mm	IVS	-	OHS	Albendazole 400 mg/day	Good, 4 mo.
Naeem S. et al. 2013 (4)	48 Female Pakistan	Shortness of breathe Loss of appetite	22x30 mm	IVS	-	OHS	Albendazole 400 mg/day	Good, 6 mo.
Inzirillo F. et al. 2013 (5)	85 Female Italy	Dyspnea	nd	Pericardial	nd	OHS	nd	Good, 6 mo.
Suner A. et al. 2014 (6)	21 Female Turkey	Dyspnea Arrhythmia (RBBB)	nd	LA, sec.ASD, PA	nd	OHS	Albendazole	Good
Yan F. et al. 2014 (7)	26 patients 11 female, 15 male Mean age:28±7,6 China	Dyspnea(15) Chest pain(11) Palpitation(8) Cough(7) Fever(2)	50x40 mm (range 10-135 mm)	LV (7) RA (2) IVS (1)	+(15)	OHS CBP(15)	Albendazole (10-15 mg/kg/day)	5 death, 1 recurrence, 20 good. 68±21 mo.
Fiergo L. et al. 2014 (8)	11 Female Turkey	Chest pain Palpitation	90x60 cm	LV	+	OHS	Albendazole 400 mg/day	Good, 18 mo.
Savaş G. et al. 2014 (9)	44 Female Turkey	Dyspnea Chest pain	53x45 mm	LV	+	OHS	Albendazole	Good, 2 mo.
Altas O. et al. 2014 (10)	3 patients (3 males) mean age:39 (24-44years)	Shortness of breathe Fatigue Chest pain	48x28, 25x21, 36x24 mm	IVS(1) LV(1) IVS(1)	nd	OHS	Albendazole 800 mg/day	Good
Yıldız CE. et al. 2014 (11)	27 Male Turkey	Dyspnea Chest pain Fatigue, Fever	100x60 mm	LV	+	OHS	Albendazole	Good, 6 mo.
Khosravi A. et al. 2014 (12)	13 Male Iran	Dyspnea Chest pain	73x53 mm	LV	+	OHS	Albendazole	Good, 2 years
Sahin I. et al. 2015 (13)	31 Female Turkey	Dyspnea Limb edema Chest pain Palpitation	55x49 cm	RV	+	OHS	Albendazole	Good, 1 year
Dasbaksi K. et al. 2015 (14)	39 Female South West Bengal	Asymptomatic	120x50 mm	LV	+	OHS	Albendazole 800 mg/day	Good, 5 years
Mirzaie A. et al. 2014 (15)	14 Male Iran	Acute lower extremity pain	40x35 mm	LV	+	OHS	Albendazole 400 mg/day	Good, 7 years
Tosya A. et al. 2015 (16)	53 Male Turkey	Recurrent arteriel embolic events	30x40 mm	Aortic lumen	nd	OHS	Mebendazole (50 mg/kg/day)	Good, 2 years
Aggeli C. et al. 2016 (17)	56 Female Greece	Asymptomatic	35x35 mm	LV	nd	No surgery	Albendazole	Good, 6 mo.
Parakh N., et al. 2016 (18)	52 Female India	Shortness of breathe Dizziness	100x80 mm	IVS	nd	OHS	nd	Exitus on the day of surgery

Jain N.et al. 2015 (19)	50 Female India	Dyspnea	78x69 mm	LV	+	OHS	nd	Good
Charfeddine S.et al. 2015 (20)	36 Female Tunisian	Chest pain Lack of appetite	50x48 mm	LV	+	OHS	Albendazole	Good, 6 mo.
Salehi R.et al. 2015 (21)	54 Female Iran	Asymptomatic	85x65 mm	IVS	nd	OHS	nd	Good
Ohri S.et al. 2015 (22)	67 Male India	Chest heaviness	23x21 mm	LV	+	OHS	Albendazole, 800 mg/day	Good, 6 mo.
Sabzi F.et al. 2015 (23)	45 Male Iran	Chest pain Dyspnea	nd	LV	+	OHS	Albendazole	Good, 6 mo.
Celik M.et al. 2016 (24)	23 Male Turkey	Exertional dyspnea	41x32 mm	RV	nd	OHS	Albendazole	Good
Saglican Y.et al. 2016(25)	35 Female Turkey	Palpitation Dyspnea	65x50 mm	IVS	nd	OHS	Albendazole	Good, 1 year
Sarli B.et al.2016 (26)	62 Female Turkey	Palpitation Dyspnea	90x60 mm	IAS	+	OHS	nd	Died of anaphylactic shock
Demirel M.,et al. 2016 (27)	55 Female Turkey	Dyspnea	nd	LV	+	No surgery	Albendazole 800 mg/day	Good, 6 mo.
Patrizia C.et al. 2016 (28)	40 Male Peruvian	Asymptomatic	nd	LV	+	No surgery	nd	Good
Mirijello A.et al. 2016 (29)	23 Male Italy	Loss of consciousness	51x43 mm	IVS	+	No surgery	Albendazole 400 mg/day	Good
Yasim A.et al. 2016 (2005-2013) (30)	25 patients 15 male, 10 female Mean:33,4±12,6 years (15-75)	Dyspnea	nd	11 intracaviter 14 extracaviter	+(22) _(3)	OHS	Nd	51±29 mo. Good, Except 1 death.
Alonso J.et al. 2016 (31)	21 Male Romanian	Loss of Memory, seizures	nd	LV	nd	OHS	Albendazole	Good
Kocabay G.et al. 2016 (32)	51 Male Turkey	Asymptomatic	nd	LV	+	OHS	Albendazole	Good
Yaman M.et al. 2016 (33)	33 Female Turkey	Chest pain During ceaseraen surgery	28x36 mm	IVS	+	No surgery	nd	Exitus
Kothari J,et al. 2016 (34)	28 Male India	Chest pain	92x96 mm	LV	nd	OHS	Albendazole, 400 mg/day	Good, 2 mo.
Sirlak M.et al. 2007 (35)	32 Male Turkey	Chest pain Weight loss Lethargy	50x45 mm	LV	+	OHS	Albendazole	Good, 1 year.

LV: Left ventricle; RV: Right ventricle; LA: Left atrium; RA: Right atrium; nd: no data; OHS: Open heart surgery; IVS: inter ventricular septum; IAS: Interatrial septum

In the following paragraph, we briefly discuss the general life cycle on the definitive and intermediate hosts. The adult forms lay eggs in the small bowel of the definitive host (commonly dog), which are passed in the feces. After ingestion by an intermediate host (sheep or goat), the egg releases an oncosphere that migrates via the circulation into different organs. The oncosphere develops

into a cyst, producing protoscolices and daughter cysts that fill the interior of cyst. The common non-urban areas' practice of feeding sheep organs to dogs in turn makes easier transmission back to the dog. Humans become infected through contact with dogs via a fecal-oral route in an incidental manner. Intermediary transmission of eggs by flies and arthropods may also result

in infection of humans [37]. The average duration of the life of cardiac echinococcosis in humans is 10–20 years and the annual growth rate of the cyst is commonly about 1–5 cm in diameter [38,39].

Primary echinococcosis of the heart represents 0,5-2% of all hydatid disease causes in endemic regions [40].

Echinococcus granulosus is widespread in regions of Eurasia, South America and Africa. In the sole animal study for the moment in literature of 39,738 domestic livestock in Pakistan, 6.67% of animals were found to be infected. The prevalence of hydatid cysts was highest in camels (17.29%) followed by sheep (7.52%), buffalo (7.19%), goats (5.48%), and cattle (5.18%) [41].

Localizations

Hydatid cysts usually affect liver and lungs, although any part of the body can be involved. These cysts can arise as part of an extensive systemic infection or as a separated event [42].

Cardiac hydatid cysts are very uncommon found in fewer than 2% of cases of hydatidosis and most usual cardiac localizations are in the myocardium of the left ventricle wall. The most common cardiac locations are the left ventricular wall (60%), followed by the right ventricle (10%), pericardium (7%), left atrium (6-8%), right atrium (3-4%), and the interventricular septum (4%) [43]. In 50% of such cardiac cases, there is multiple organ inclusion [44].

In this review of literature, 25 cysts were in the left ventricle, 3 were in the right ventricle, 11 were in the interventricular septum, one was in the aortic lumen and others were extended to multiple localizations. All serologic tests were positive except three of them and the ones that were not given data.

Larvae usually extend to the myocardium through the systemic or pulmonary circulation or as direct extension from near organs and that is a uncommon localisation.

Clinical presentation

The clinical presentation in cardiac hydatid cyst is commonly spreading in a hidden way. The slowly growing larger echinococcal cyst generally remains asymptomatic until the space-occupying effect in an included organ brings out symptoms. Early diagnosis and management are very significant given the lethal risk of cyst perforation. The endocardial infection occurs infrequently in comparison to the myocardial involvement. Again, hydatid cysts may seldom develop within pulmonary arteries following ruptured cardiac or hepatic cysts [45].

The disease can stay asymptomatic (90%), but it may cause change of position of the coronary vessels, arrhythmias and mechanical interference with the atrioventricular valves and ventricular function via cysts' mass effect. Compression of the coronary arteries by a cyst can cause myocardial infarction. Pulmonary embolism could complicate the course. It may result from rupture of cardiac or hepatic cysts and may only be diagnosed postmortem [46-50].

Common symptoms were dyspnea, chest pain, palpitation, loss of appetite and fever. Two patients had acute peripheral arterial embolic events and two had cerebral events with loss of consciousness and seizures.

Cardiac echinococcosis can be manifested by rhythm disturbances, angina pectoris, valvular malfunction, pulmonary or systemic embolism, and right or left ventricular outflow tract obstruction. Cyst perforation is the most hazardous complication, ending in anaphylactic shock, thrombo embolism, and death in approximately 75% of patients [51,52].

DIAGNOSIS

The diagnosis of cardiac echinococcosis can be hard and must be suspected in any patient from endemic or sheep farming areas with a cystic tumour of heart. The diagnosis is usually achieved by combination of clinical findings, imaging and serology. For radiological image part of cardiac echinococcosis, chest X-ray, echocardiography, computerized tomography (CT) or magnetic resonance imaging (MRI) might be used.

Transthoracic echocardiography is showing the cyst with echo-negative components and regular contours is the most effective method of diagnosing the hydatid cyst. It is possible to get more detailed images with transoesophageal echocardiography. Echocardiography is the preferred diagnostic method because of its low cost and availability. Nevertheless, echocardiography is operator-dependent, with a limited field of view to see the area behind the sternum may be difficult or impossible to examine [53].

Other diagnostic steps to be taken accordingly enclose CT scan and MRI, and the performance of serologic tests. CT and MRI are superior to echocardiography for evaluation of pericardial masses and their relationship to surrounding tissues and extension to myocardium. CT can identify cysts, but it often fails to detect small lesions, it best shows wall calcification. MRI is the technique chosen for follow up. Specific signs of hydatid cyst include calcification of wall, the presence of daughter cysts, and membrane detachment [54].

Routine laboratory tests are not specific and may reveal normal or abnormal values. The blood count may show eosinophilia. Serologic tests are of limited value in diagnosis. The Caroni intradermal test and serology (indirect hemagglutination in (IHA) or enzyme-linked immune sorbent assay (ELISA) techniques) are useful but are subject to false negativity [55].

ELISA has sensitivity and specificity of 94% and 99%, respectively, and along with IHA test, it has proved to be the best choice for follow-up. Indirect fluorescent antibody and enzyme immunoassay complement fixation (Weinberg) tests are also used [56].

In patients with initial negative serology, serotesting using IHA, latex agglutination or radioallergosorbent testing should be done as a combination of several tests can improve the diagnostic exactness [57].

TREATMENT

Surgery has customarily been the primary method of treatment due to the high risk of associated complications, even in asymptomatic cases, and the safest way to remove the cyst definitely is under cardiopulmonary bypass. The principles of surgery are to avoid handling of heart before cross clamping, and complete excision of germinal layer. Some prefer to let the

residual cavity heal by secondary intent to avoid complications. Preventing contamination of the surgical field is extremely important [58]. Surgical risks include anaphylaxis and dissemination due to the cystic fluid.

Multiple agents have been intended as helminthicides: 2% formalin, 0.5% silver nitrate solution, 20% hyper tonic saline solution, 1% iodine solution, or 5% cetrimonium bromide solution. There is no evident superiority and preference among these substances. It has been suggested that hypertonic saline solution-soaked pads be distributed within the pericardial cavity in order to prevent dissemination intra operatively. Albendazole, an active agent against Echinococcus, should be administered adjunctively. It is cautious to begin albendazole several days before surgery and continue for several weeks after resection [59-61]. About 10% of all hydatid cysts tend to occur again after surgery, but may decrease with convenient medical treatment [62,63]. To reduce the recurrence, supplemental medical treatment with mebendazole or albendazole is recommended. Chemotherapy is contraindicated for large, inactive or calcified cysts with a risk of rupture [64]. There are also few systemic conditions that cause contradiction of pharmacotherapy as bone marrow depression and pregnancy. Prophylactic chemotherapy of albendazole duration can be regulated according to the number of cystic lesions and organ extension [65].

As conclusion, cardiac hydatidosis should be considered in the differential diagnosis of tumoral and cystic masses and treat properly with combination of different methods.

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