

A Case of Multiple Bilateral Pulmonary and Intracardiac Hydatidosis

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ABSTRACT

Hydatid cyst disease (HCD) is a parasitic infection caused by *Echinococcus granulosus* larvae. Habitat of the disease is most frequently liver, followed by lungs; intracardiac occurrence is rare. When it occurs, cardiac involvement is more common in left ventricle, where blood supply is intense. Disease is extremely rare in right atrium. While following case at clinic with diagnosis of bilateral pulmonary hydatid cyst, right atrium involvement was incidentally found. Since hydatid cyst disease with intracardiac localization is rarely seen, present report was prepared in order to draw attention to importance of thorough diagnosis and treatment of these patients.

INTRODUCTION

Hydatid cyst disease (HCD) is zoonosis frequently caused by *Echinococcus granulosus* (EG), and encountered as endemic disease in some geographic regions, including Turkey. Its annual incidence has been reported by Cangir et al. as 12/100,000, and it continues to be an important health problem, especially in rural areas.^[1] Human beings are infected when eggs of the parasite expelled with stool of canidae, the definitive hosts during life cycle of the parasite, are ingested. Most frequently (65%), liver is affected. Orally ingested parasite eggs are absorbed by intestinal system, and via portal circulation, embryos typically settle in liver and develop into cystic metacestodes. In approximately 10–30% of cases, parasite larvae pass through hepatic sinusoids and reach lung tissue via vena cava inferior and pulmonary artery. Parasites that pass into systemic circulation without being retained in capillary system of these 2 organs can infect any tissue or organ of the body.^[2–4] HCD caused by EG in intermediate host, i.e., human beings, rarely affects heart (0.5–2%), and when it does,

most often left ventricle is involved.^[5] Hydatid cysts may result in highly variable and serious symptoms, according to location in heart. They can cause cardiac rupture, leading to fatal outcome due to cardiac tamponade, anaphylactic shock or systemic embolization.^[6] Therefore, surgery should be considered when possible even for patients who are asymptomatic at time of diagnosis of intracardiac involvement.

CASE REPORT

A 25-year-old male patient presented to clinic with exertional dyspnea, chest pain, bloody sputum, and testicular swelling. Physical examination findings were as follows: moderately good general health, open conscious, full cooperation and orientation, body temperature: 36.5 °C, arterial blood pressure (ABP): 110/70 mm Hg, RR :21/min Left lower quadrant rales were detected during respiratory system examination. Cardiac examination revealed physiological first (S1) and second (S2) heart sounds, as well as third heart sound (S3), without any additional

murmur. Liver was palpable 2–3 cm below costal margin. There was eruption on patient's back that did not fade with pressure. On urogenital system examination, right testis was found to be minimally larger than left without any palpable mass.

Laboratory values were as follows: White blood count (WBC): 9900/uL, platelet (PLT): 212000/uL, hemoglobin (Hb): 13.7g/dL, hematocrit (Htc): 41.1%, erythrocyte sedimentation rate (ESR): 21 mm/h, C-reactive protein (CRP): 10 mg/L, blood urea nitrogen (BUN): 36 mg/dL, creatinine 1.02 mg/dL, alanine aminotransferase (ALT): 28 U/L, aspartate aminotransferase (AST): 29 U/L, lactate dehydrogenase (LDH): 438 U/L, sodium (Na): 140 mEq/L, potassium (K): 5.18 mEq/L, calcium (Ca): 8.9 mEq/dL, chloride (Cl): 109 mEq/L, and coagulometry and urine analysis results were within normal limits.

On posteroanterior (PA) chest radiograms, multiple non-homogeneous opacities with regular contours were seen bilaterally in all zones. On non-contrasted thoracic computed tomograms (CT), lymphadenopathies of pathological size were observed on right upper and lower paratracheal regions of mediastinum, right hilar region, and bilateral parenchymal nodules were reported. Regular contours were widespread on all lobes with similar characteristics, and size suggested prior metastatic involvement (Figure 1, 2). Respiratory function tests revealed FEV1/FVC: 85%, FEV1: 2.10 lt (54%) FVC: 2.49 lt (54%). Oxygen saturation in room air was 97%. Relevant tumor markers were within normal limits, while hydatid cyst indirect hemagglutination assay (IHA) (1/2048) positivity was found. Urinary system and scrotal ultrasound results were within physiological limits. Urology consultation

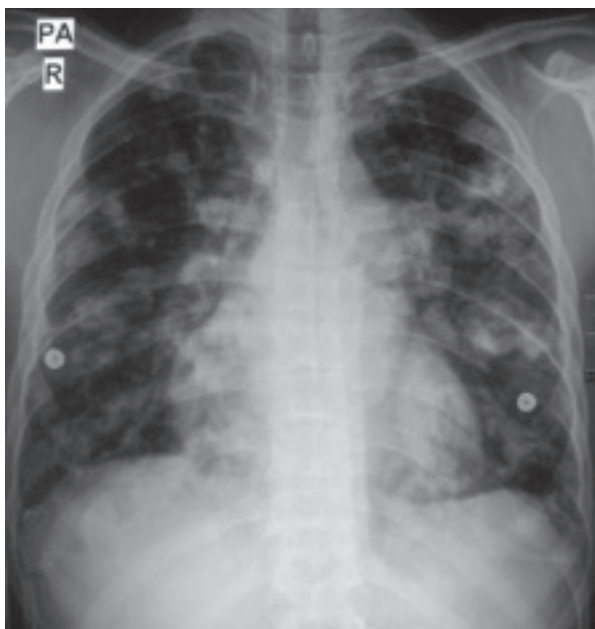


Figure 1. Chest X-ray obtained at admission.

was performed, and no urological pathology was detected. On abdominal ultrasound (US), no significant lesion was noticed regarding hepatic hydatid cyst; however, on thoracic US, a suspect lesion was seen on wall of right atrium, which prompted echocardiography (ECG) request (Figure 3). On echocardiograms, following images were observed: exceedingly enlarged right heart chambers, pulmonary artery pressure: 100 mm Hg, ejection fraction (EF): 65%, mass lesion measuring 3x3 cm in right atrium originating from free wall of right atrium. Thoracic CT angiograms of patient were obtained and revealed non-contrasted hypodense area on wall of right atrium, necessitating cardiovascular consultation (Figure 4, 5). Cardiovascular surgeon evaluated patient and decision was made for surgical intervention. Patient was transferred to

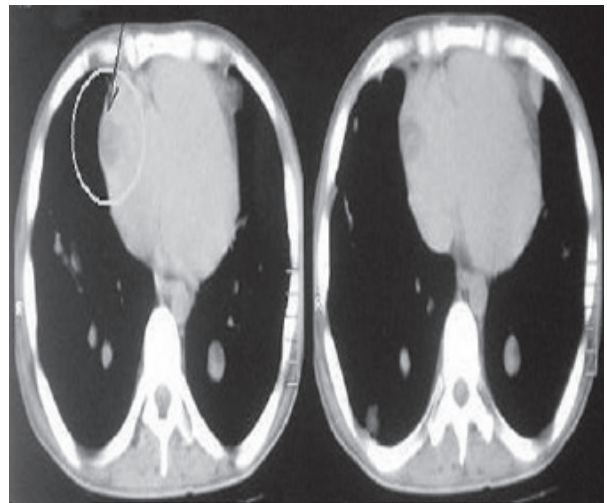


Figure 2. Thoracic CT image of cyst localized in the right atrium.

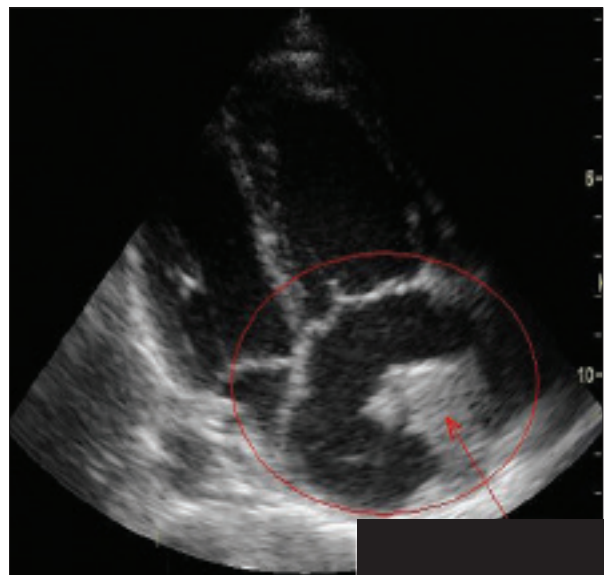


Figure 3. ECG image of the cyst localized in the right atrium.

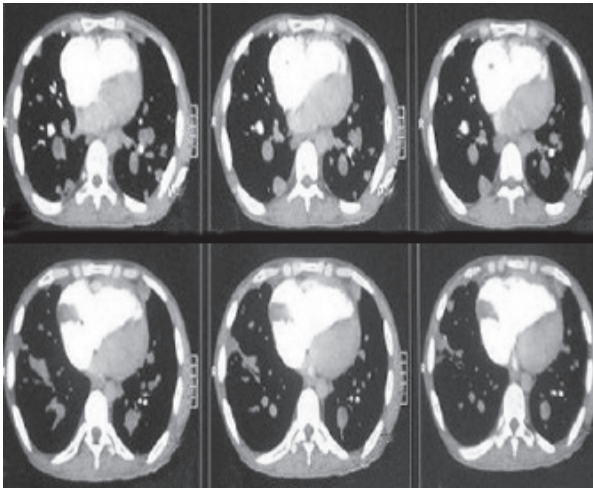


Figure 4. Thoracic CT angiographic sections from mediastinal window.

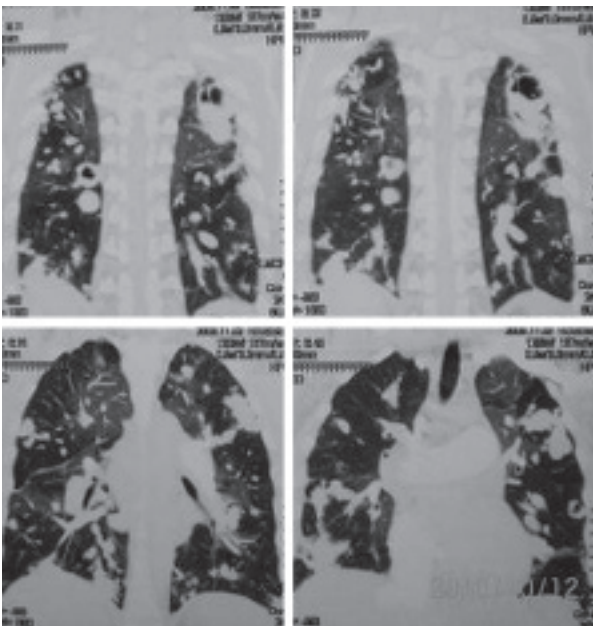


Figure 5. Thoracic CT angiographic sections from parenchymal window

cardiovascular surgery clinic and underwent cardiac surgery with initial diagnosis of HCD and atrial mass was removed. Postoperative pathology report indicated hydatid cyst. Albendazole treatment was initiated.

DISCUSSION

Pathogenic agent of EG is cestode larvae, and most pulmonary infestations are caused by EG. Adult worm is composed of 4 segments: head (scolex) and three proglottids. It has 4 suckers on its head, and rostellum contains hooks that attach to intestinal mucosa. Gravid proglottid carries the eggs of the parasite. Adult mature worms live in the

guts of the definitive host (canines). Eggs are ejected into environment with stool of definitive host. Parasite eggs taken via oral route by intermediate hosts (e.g., human beings, sheep, cow or pig) develop into larvae in duodenum. These larvae penetrate intestinal wall, enter portal blood circulation, and reach the liver, where they usually attach themselves to sinusoids. Some larvae advance beyond hepatic sinusoids and are retained in alveolar capillaries. Most larvae retained in organs die; however, a few cause infestation.^[7,8]

Since cardiac contractions form a natural resistance, cardiac hydatid disease is rarely seen. Hydatid cysts settle in the heart (0.02–2%), left (55–75%) and right (15–18%) ventricles, septum (5–9%), and right atrium (3–4%) in indicated percentages. In a series of 6 cases, Aytaç et al. reported presence of hydatid cysts in left ventricle of 5 patients and right ventricle of 1 patient.^[9] In present case, intracardiac cyst, the rarest form of hydatid cyst, was localized in right atrium. Embryos of parasite enter heart through pulmonary or coronary artery, and adventitial pericyst layer forms as myocardial reaction against presence of cyst. The most frequently encountered clinical signs of intact cardiac hydatidosis are precordial chest pain and coughing; however, when cyst wall ruptures, fever, hemoptysis, dyspnea, anaphylactic shock, syncope, arrhythmia and conduction disturbances, acute myocardial infarction, pericarditis, valvular dysfunction, pulmonary hypertension, pulmonary or systemic embolism, and sudden death can occur. Present patient had no cardiac pathology and only minimal complaint. Therefore, it should not be forgotten that patients with cardiac hydatid cyst are often asymptomatic.^[10,11]

Although cardiac HCD is generally asymptomatic, since serious complications can occur, and rupture of cyst wall may cause sudden death, definitive treatment for cardiac hydatid cyst is surgery. Surgical mortality for cardiac hydatid cyst reportedly ranges between 0.29 and 0.6%. Clinical entity manifests itself more frequently according to size and location of cyst and related complications.^[12,13]

HCD is still widely seen in Turkey. Although it most often affects liver, many tissues and organs can become infected. In particular, as seen in present case, it should be remembered that intracardiac cysts can be found in cases with bilateral and multiple pulmonary involvement. Since intracardiac cysts are generally asymptomatic, these cases should be scanned with ECG, which is a simple, inexpensive, and reliable diagnostic method. Although intracardiac cyst usually leads asymptomatic course, since there can be fatal outcome, when possible, surgical intervention should be considered for patients diagnosed as cardiac hydatid cyst.

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İntrakardiyak ve İki Taraflı Pulmoner Multipl Hidatidoz Olgusu

Kist hidatik hastalığı, *Echinococcus granulosus*'un larvasının etken olduğu parazitik bir enfeksiyondur. Yerleşim sıklığına göre en çok karaciğer ve daha sonra akciğerde görülen hastalıkta intrakardiyak yerleşim nadir görülmektedir. Kardiyak yerleşim kanlanmanın daha çok olduğu sol ventrikülde daha sıktır. Sağ atriyum yerleşimi en nadir görülen formudur. Kliniğimizde iki taraflı pulmoner kist hidatik tanısı olan ve bu tanı ile takip edilirken rastlantıyla intrakardiyak (sağ atriyum) yerleşim tespit edilen bir olguyu intrakardiyak yerleşimli kist hidatik hastalığının nadir olması ve bu hastalarda tanı ve tedavinin önemine dikkat çekmek için yayınlamayı uygun bulduk.

Anahtar Sözcükler: Kist hidatik; intrakardiyak kist hidatik.