

Cystic Echinococcosis: One Entity, Two Unusual Locations

Kistik Ekinokokkoz; Bir Hastalık, İki Sıra Dışı Yerleşim

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ABSTRACT

We report two cases of cystic echinococcosis at unexpected locations. Patients were a 64-year-old man and 35-year-old woman. A cystic mass was incidentally found between the prostate and seminal vesicles in the male patient, using ultrasonography during a check-up for ischemic coronary disease. The female patient was admitted to the hospital with symptoms of cardiopulmonary disease, but her detailed radiological examination showed a cystic lesion in the interventricular septum of the heart. Both patients were operated, and examinations of the histologic sections revealed cyst walls consistent with echinococcal infection. Attention should be focused on this entity even in endemic areas, and pathologists should be aware of the histologic characteristics of this lesion, to avoid misdiagnosis as a nonspecific cyst. (*Türkiye Parazitol Derg* 2016; 40: 51-3).

Keywords: Echinococcosis, hydatid cyst, prostate, heart

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ÖZ

Bu makalede, biri 64 yaşında erkek, diğeri 35 yaşında kadın olan iki ayrı hastada sıra dışı yerleşim gösteren tek bir hastalık, kistik ekinokokkoz sunulmaktadır. Erkek hastanın iskemik koroner hastalığı nedeniyle yapılan genel kontrolleri sırasında, ultrasonografi sonucu prostat ve vezikula seminalisler arasında kistik kitle saptanmıştır. Diğer hasta ise kardiopulmoner şikâyetleri nedeniyle hastaneye başvurmuş, görüntüleme yöntemleri ile kalp ventrikülleri arasında kistik bir lezyon belirlenmiştir. Her iki hastada lezyonlar rezekt edilmiş, histolojik inceleme sonucu kistik ekinokokkoz enfeksiyonu tanısı verilmiştir. Endemik bölgelerde alışlagelmiş lokalizasyonlarda sıklıkla görülmekle birlikte, bu kistlerin sıra dışı yerleşim gösterebileceği ve bu nedenle spesifik olmayan kistlerle karışabileceği akılda tutulmalıdır. (*Türkiye Parazitol Derg* 2016; 40: 51-3).

Anahtar Kelimeler: Ekinokokkoz, kist hidatik, prostat, kalp

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INTRODUCTION

Cystic echinococcosis (hydatid cyst) is a parasitic disease caused by infection with the larval stage of *Echinococcus granulosus*, a 2-7 millimeter long tapeworm found in dogs (definitive host) and sheep, cattle, goats, and pigs (intermediate hosts). Humans are infected by the ingestion of parasite eggs through contaminated food or water or through a direct contact with animal hosts. Most infections in humans

are asymptomatic for many years until cysts grow to an extent that triggers clinical signs.

Cystic echinococcosis in endemic areas is common in routine surgical pathology practice. In general, the cysts are most commonly located in the liver and lungs. They are usually diagnosed under detailed pathologic evaluation and clinicopathologic correlation. However, in lesions mimicking hydatid cysts, particularly in unusual sites, both the

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imaging techniques and histopathologic examination can lead to inappropriate differential diagnosis. Here we present two cases of cystic echinococcosis with their clinical and histopathologic appearance and a brief literature review.

CASE REPORT

Case 1

A 64-year-old man was followed up for ischemic coronary disease. During the last consultation, a well-limited, 65-mm mass composed of cystic and solid components at the posterior of the urinary bladder was detected using abdominal ultrasonography. Its association with prostate or seminal vesicle was not well defined using ultrasonography. Both urinary system and transrectal ultrasonography showed a cystic and semisolid mass measuring 155 mL in volume that was closely associated with prostate and seminal vesicles. Initial diagnosis was done using trucut needle biopsy and aspiration of the cyst fluid. Microscopically, the sections showed fibromuscular prostatic tissues and isolated thick fibrous cyst wall fragments without any cellular lining and dispersed multinuclear histiocytic giant cells in the wall (Figure 1). Cytospin preparations of the cyst fluid showed proteinous secretion, nuclear debris, and spermatozoa. The diagnosis was benign cyst wall consistent with either a prostatic cyst or seminal vesicle cyst.

Based on these findings, transurethral prostatectomy was performed. The resected tissues were 23 gm in weight and 6.5x6x0.6 cm in dimensions. Along with the prostatic tissues were 4x3.5x0.5 cm pearly membranous tissues. Microscopic sections revealed cellular, lamellous cyst wall fragments consistent with cuticula between the prostatic tissues, but no scolex was detected (Figure 2).

Case 2

A 35-year-old female presented with palpitation and dyspnea. Echocardiography and cardiac MRI revealed a 5-6 cm interventricular septal cardiac cyst. The patient was prepared for surgery after a preoperative diagnosis of hydatid cyst. At the beginning of surgery, transesophageal echocardiography was performed to verify the localization of the lesion; it was located at the septum, bulging toward the posterior wall of the ventricle. The cyst fluid was aspirated, and the wall was resected from the septum.

On gross examination, the specimen consisted of membranous calcified tissues and cuticular membrane. Histologic sections revealed, a diffusely calcified and inflamed fibrous capsule containing residual muscle of the heart at the outer portion. There were acellular, laminated chitinous membrane and degenerated germinal layer at the inner wall of the fibrous capsule (Figure 3). A few scolices were also detected (Figure 4).

Outcome

Both the lesions were diagnosed as cystic echinococcosis; one of them in the prostate and the other in the heart. Anti-Echinococcus total antibody was found to be elevated on serologic examinations, and allergic laboratory tests showed high levels for specific anti-Echinococcus IgE antibodies in both patients. Other organs were scanned to locate the primary site of the disease or to identify haematogenous spread of Echinococcus. A calcified cystic lesion consistent with hydatid cysts was found in the liver of case 1; however, results from the imaging techniques were within normal limits in case 2 and no other focus was detected. Albendazole was used after surgery,

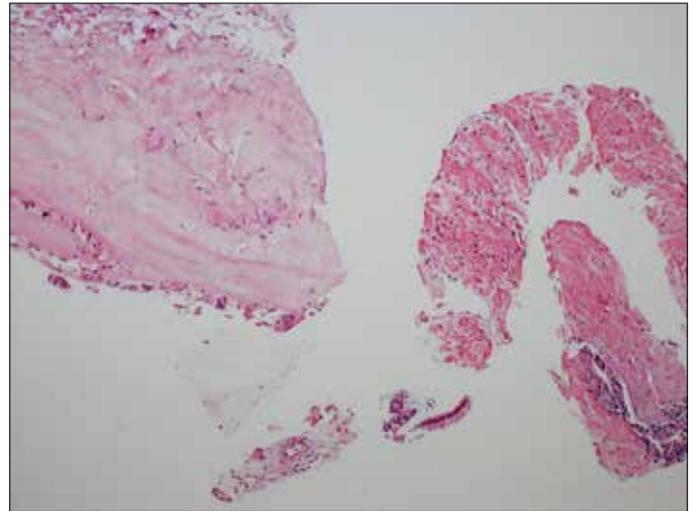


Figure 1. Trucut needle biopsy revealed prostatic tissues and isolated thick fibrous cyst wall fragments (H&E, x100)

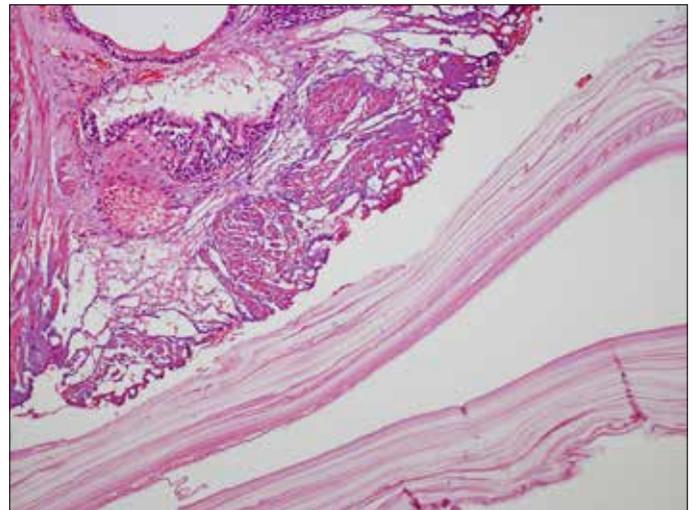


Figure 2. Transurethral resection specimen of the prostate; prostate tissue (left) and a cellular, lamellous cyst wall (right) (H&E, x100)

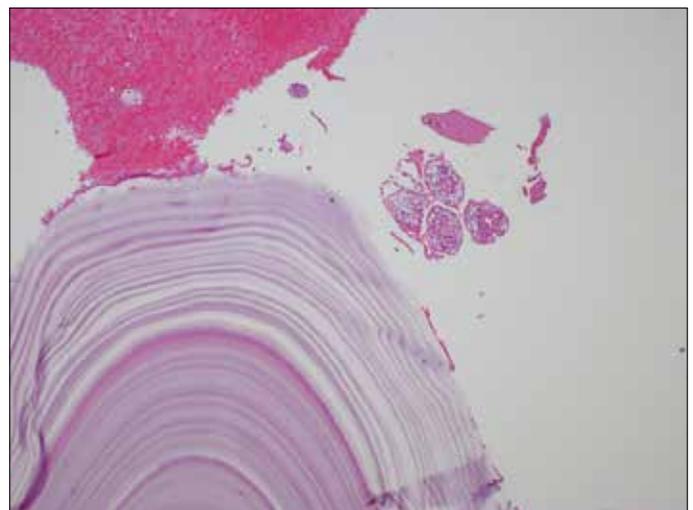


Figure 3. Cardiac cyst wall consists of a laminated chitinous membrane and few scolices (H&E, x100)

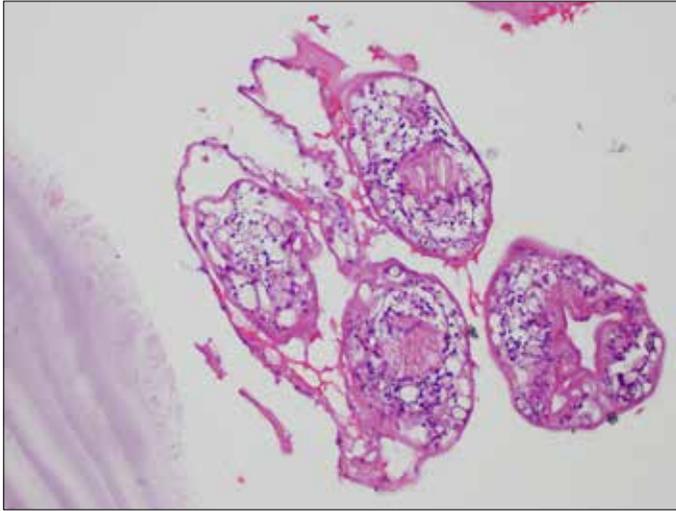


Figure 4. Scolices within the cyst (H&E,×200)

and there was no evidence of the hydatid disease at their post-operative 1 year follow-up.

DISCUSSION

Echinococcal infection is caused by hydatid tapeworm, mostly *E. granulosus* (99%) (1). It is a common disease particularly in the Middle East, Greece, Australia, North Africa, and some parts of South America (2). It may occur at any age and most commonly involves the liver (50%-70%) followed by the lungs (11%-17%) (1, 2). Compared with the heart, prostatic or seminal vesicle hydatid cysts are less common (3, 4). Davies (5) referred to the history of cystic echinococcosis and related it to the time of Hippocrates, but probably the early cases related to the prostate were reported by Plaggmeyer and Cumming in 1922. Subsequently, more cases were reported that included the kidney, spleen, urinary bladder, bone, retrovesical, and spinal cysts (4, 6-9).

Clinical manifestations of hydatid disease were variable, and some of them were incidentally detected during radiologic investigations. Ultrasonography and computerized tomography (CT) cannot always reliably distinguish these lesions from other cystic lesions, such as congenital müllerian remnant cysts, retention cysts, and even cystic degeneration of prostatic hyperplasia. Differential diagnosis should include simple cysts, cystadenomas, and teratoma. Tumors and aneurysms of the heart should also be considered. Causes of diagnostic mistakes using CT include early disease stage, small cyst size, features of localization, and lack of daughter cysts. Cysts can be primary in these locations, but in published reports, they are mostly believed to have an origin from other usual locations (1). Serologic findings are about 90% sensitive for *E. granulosus*; however, false-positive results do occur. Definitive diagnosis is based on cyto-histologic findings; a meticulous search for acid-fast birefringent hooklets or laminated membrane is mandatory.

Cystic hydatid disease has generally been treated with surgical resection. The introduction of other interventional methods, such as percutaneous drainage, combined with albendazole therapy is an effective alternative treatment, particularly for hydatid cysts of the liver. Surgery with the pre- and post-operative administration of albendazole is the best treatment option (10).

CONCLUSION

An awareness of the existence of these lesions even in endemic areas and a careful focus on key histologic characteristics will aid pathologists in appropriate diagnosis.

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