original article

# Clinical characterization of unusual cystic echinococcosis in southern part of Turkey

Atılgan Tolga Akcam,[a] Abdullah Ulku,[a] Ismail Soner Koltas,[b] Volkan Izol,[c] Omer Sunkar Bicer,[d] Emine Kilicbagir,[e] Gurhan Sakman,[a] Hakan Poyrazoglu,[f] Tahsin Erman,[g] Ibrahim Atilla Aridogan,[c] Cem Kaan Parsak,[a] Mehmet Inal,[h] Serdar Iskit[i]

From the [a]Department of General Surgery, [b]Department of Parasitology, [c]Department of Urology, [d]Department of Orthopedics, [e]Department of Pathology, [f]Department of Cardiac Surgery, [g]Department of Neurosurgery, [h]Department of Radiology, and [i]Department of Pediatric Surgery, Cukurova University, Adana, Turkey

Correspondence: Ismail Soner Koltas, PhD · Department of Parasitology, Cukurova University, Balcali, Adana 01330, Turkey · T: +903223386060 F: +903223386572· koltas@cu.edu.tr

Ann Saudi Med 2014; 34(6): 508-516

DOI: 10.5144/0256-4947.2014.508

**BACKGROUND AND OBJECTIVES:** The incidence of primary extrahepatic cystic echinococcosis (CE) is rare. Generally, radiological and serological findings can help establish the diagnosis of hepatic and pulmonary CE, but a CE in an unusual location with atypical radiological findings may complicate the differential diagnosis. The objective of this study is to present the characteristics of cases with extrahepatic CE in respect of sites of involvement, clinical presentations, radiological findings, serological diagnostic evaluations, and outcomes of infected patients.

**DESIGN AND SETTINGS:** A retrospective analysis of surgically treated CE was conducted between January 1993 and January 2014 in the General Surgery, Pediatric Surgery, Urology, Cardiovascular Surgery, Neurosurgery, and Orthopedics departments of University of Cukurova, Faculty of Medicine, Balcalı Hospital.

**PATIENTS AND METHODS:** Among the 661 patients managed for CE, 134 had unusual sites of involvement. Radiological and serological examinations were used to differentiate CE from alveolar echinococcosis.

**RESULTS:** Of 134 cases with unusual sites of involvement, 32 cases had liver CE (23.9%), 7 cases had lung CE (5.2%), and 2 cases had concomitant liver and lung CE (1.5%). In 93 (69.4%) cases, unusual organ involvement was isolated without any liver or lung involvement. The mean age was 45 years. Abdominal pain was the main symptom and was found in 104 patients. Thirty-one (23.1%) of 134 extrahepatic CE cases were evaluated as negative with indirect hemagglutination (IHA). However, positive results were obtained in 54 cases evaluated with Echinococcus granulosus IgG Western blot (WB), including 10 IHA-negative cases.

**CONCLUSION:** CE with unusual localizations may cause serious problems of diagnostic confusion. The combination of clinical history, radiological findings, and serological test results (especially the WB) are valuable in diagnosing extrahepatic CE.

Cystic echinococcosis (CE) is endemic in the Mediterranean region, Central Asia, the Middle East, East Africa, Australia, New Zealand, and the southern part of South America.[1-3] Human CE infections occur by accidental ingestion of eggs excreted with feces of infected carnivores. Oncospheres hatch in the duodenum, penetrate the intestine, and are transported via the bloodstream to various organs. The liver is the most commonly involved organ (52%-77%) because the liver filters oncospheres (if any) from the blood that flows from the portal vein, which drains the intestinal veins.[4] If oncospheres escape this filtering process, they are introduced into the systemic circulation and settle in the lungs (10%-40%) or, rarely, in other organs (10%). Extrahepatic and extrapulmonary locations of the disease are described as "unusual" in previous articles.[4,5] Because the symptoms vary according to the organ localization and the rate of parasite growth, the clinical diagnosis of CE is not easy. Medical radiological findings such as ultrasonography (US) is the first choice in the diagnosis of intra-abdominal cysts because US is easy to use and effective, and it is considered to be the "gold standard" in community-based surveys. Serology gives useful and complementa-

ry information on the nature of the cyst and concerning the issue of surgical intervention.[5-10] Early diagnosis and subsequent treatment can reduce mortality. The risk of postsurgical relapses is one of the problems that can be faced after treating CE patients.[11-13]

In this article, we described the results of 134 patients in respect of sites of involvement, clinical presentations, radiological findings, serological diagnostic evaluations, and outcomes of infected CE patients..

## PATIENTS AND METHODS

A retrospective analysis of surgically treated CE was conducted between January 1993 and January 2014 in the General Surgery, Pediatric Surgery, Urology, Cardiovascular Surgery, Neurosurgery, and Orthopedics departments of University of Cukurova, Faculty of Medicine, Balcalı Hospital. Among the 661 patients managed for CE, 134 had unusual sites of involvement, and these patients constituted the population of this retrospective study.

To diagnose and determine the extent of the disease, US, computed tomography (CT), magnetic resonance imaging (MRI), conventional x-ray, intravenous urography, and transesophageal echocardiography (TEE) or transthoracic echocardiography (TTE) were used, and the modality chosen depended on the site of involvement. Generally, US and/or CT were the methods of choice in cases of abdominal involvement, while chest x-ray (CXR) and/or CT were used in cases of thoracic involvement. CT, MRI, and/or TEE were used in the case of cardiac disease. In cases of central nervous system (CNS) and spinal involvement, evaluators used CT and/or MRI; in cases where bones were involved, x-ray and/or MRI were preferred.

For the serological evaluation, the indirect hemagglutination (IHA) test was performed in all cases; however, a Western blot (WB) was performed in the 54 cases that occurred since the year 2000. Anti-*Echinococcus granulosus* antibodies were determined using IHA (Fumouze Diagnostics, France) and IgG WB (Euroimmun, Germany) techniques, each according to the manufacturer's instructions. The diagnostic value for IHA was accepted as ≥1:320 titration; for WB, the presence of a p7 band was accepted as diagnostic.

Radiological methods and serological tests were used in the postsurgical follow-up of all patients.

## RESULTS

There were 58 men and 76 women, with ages ranging from 6–67 years (average 45 years). Approximately 20.3% (134/661) of patients had cysts in an uncommon location. The results of uncommon locations of CE in the southern part of Turkey are given in **Table 1**. Among the 134 patients who had an unusual site of involvement, 41 cases (30.6%) also had liver (32 cases), lung (7 cases), or concomitant liver and lung (2 cases) involvement. In 93 cases (69.4%), unusual organ involvement was isolated without any liver or lung involvement.

### *Peritoneal cavity*

The 24 patients with peritoneal CE underwent surgery. Among the 16 patients with isolated peritoneal disease,

**Table 1.** The results of uncommon locations of cystic echinococcosis in the southern part of Turkey.

| Site | Number | % | IHA (-/+) | WB (-/+) | X-ray | US EKO | CT | MR | Liver | Lung | Recurrence |
|---|---|---|---|---|---|---|---|---|---|---|---|
| Peritoneal cavity | 24 | 3.6 | 19/24 | 2/2 | - | 21 | 24 | - | 6 | 2 | - |
| Retroperitoneum | 12 | 1.8 | 9/12 | 5/5 | - | 12 | 12 | - | 3 | 1 | 1 |
| Kidney | 18 | 2.7 | 11/18 | 7/7 | - | 18 | 18 | 5 | 2 | - | - |
| Spleen | 16 | 2.4 | 13/16 | 7/7 | - | 12 | 16 | - | 5 | 1 | - |
| CNS | 16 | 2.4 | 14/16 | 6/6 | - | - | 16 | 4 | 5 | 3 | - |
| Muscle | 14 | 2.1 | 10/14 | 5/5 | 9 | 9 | 12 | 11 | 3 | - | - |
| Bone | 14 | 2.1 | 12/14 | 6/6 | 14 | 14 | 14 | 10 | 4 | - | 2 |
| Heart | 13 | 1.9 | 11/13 | 6/6 | 13 | 12 | 12 | 10 | 4 | 2 | - |
| Pancreas | 3 | 0.5 | 2/3 | 1/1 | - | 3 | 3 | 3 | - | - | - |
| Ovary | 2 | 0.3 | 2/2 | 2/2 | - | 2 | 2 | - |  | - | - |
| Thyroid | 1 | 0.2 | 1/1 | - | - | 1 | 1 | - | - | - | - |
| Breast | 1 | 0.2 | 1/1 | - | - | 1 | - | - | - | - | - |

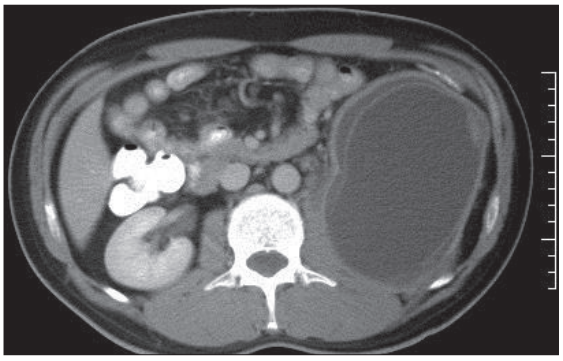

**Figure 1.** Large retroperitoneal cystic echinococcosis seen on upper abdominal CT image.

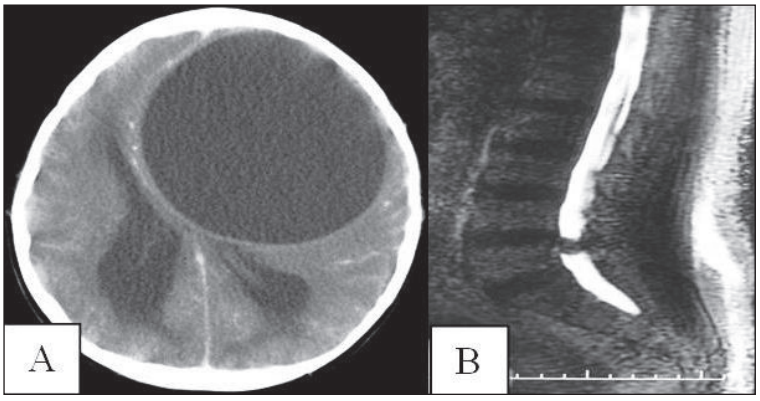


**Figure 2.** A) CE occupying left frontal lobe of brain. B) CE of spinal cord.

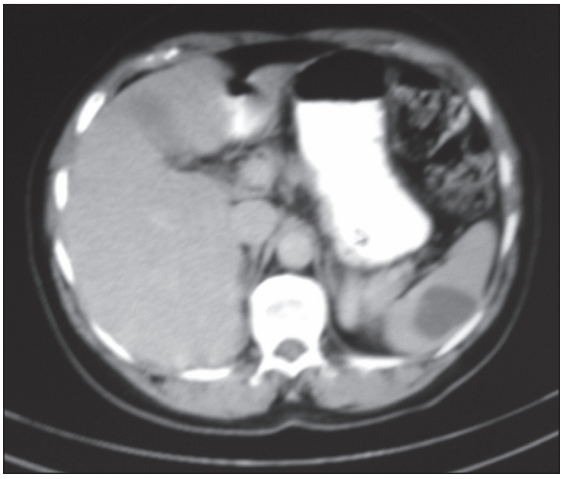

**Figure 3.** The tomographic image of CE of spleen.

5 had a history of a liver or a lung surgery for hydatidosis, though at the time of the peritoneal disease, they were free of lung or liver disease. The IHA test was positive in 19 of 24 (79.2%) cases. The WB serological test was positive in 10 cases. Among the 5 IHA-negative cases, the WB test was positive in 2. During the follow-up of 19 cases (median follow-up: 60 months), no recurrence was detected.

*Retroperitoneum*

Twelve patients had retroperitoneal disease involvement. The most frequent presenting symptoms were abdominal pain and discomfort. Three patients had undergone previous surgeries for hepatic CE. CT demonstrated all of the lesions in all patients, whereas US did not detect any lesions in 2 of the patients (**Figure 1**). The IHA test was positive in 9 (75%) cases. The WB test was performed in 5 cases, and all 5 were positive, though one of these was IHA negative. During follow-up, 1 patient who had previously undergone drainage of multiple lesions had a recurrence in 1 lesion in the third postsurgical year. The recurrent lesion was treated percutaneously with the PAIR (puncture, aspiration, injection, and re-aspiration) technique,[4] and no recurrence was detected during the 5-year follow-up period.

*Central nervous system*

Sixteen patients presented with neurological symptoms caused by CE of the CNS. Ten the patients were children (63%) (median age 8 years) and 4 were adults (37%). The most frequent presenting clinical symptoms and signs were headache (83%), papilledema (64%), and focal neurological deficits (33%). None of the patients had a history of surgery for CE. In 15 patients, lesions were located in the brain (**Figure 2A**), while 1 patient had a lesion located in the spinal canal (**Figure 2B**). The IHA tests were positive in 14 patients (87.5%). The WB test was performed in 6 cases, and positive results were obtained in all of them. During the follow-up period (median follow-up: 60 months), no recurrence was detected.

*Spleen*

Patients with CE of the spleen presented with nonspecific symptoms such as left upper abdominal pain (80%) and splenomegaly (60%). However, in 1 case, repeated gastric variceal bleeding due to segmental portal hypertension was the first symptom. The spleen was infected with CE in 16 patients (**Figure 3**). Four of these patients had undergone previous surgeries for hepatic (3 patients) or pulmonary (1 patient) CE. Involvement was isolated in the spleen in 10 (63%) cases, and concomitant liver lesions were detected in 6 (37%) cases. The IHA test was positive in 13 cases (81%). The WB test was performed in 7 cases; positive results were obtained in all cases, though IHA results were negative in 2 cases.

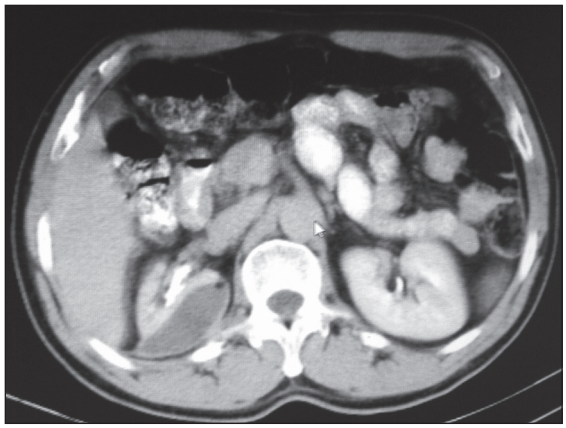

**Figure 4.** The tomographic image of CE of right kidney.

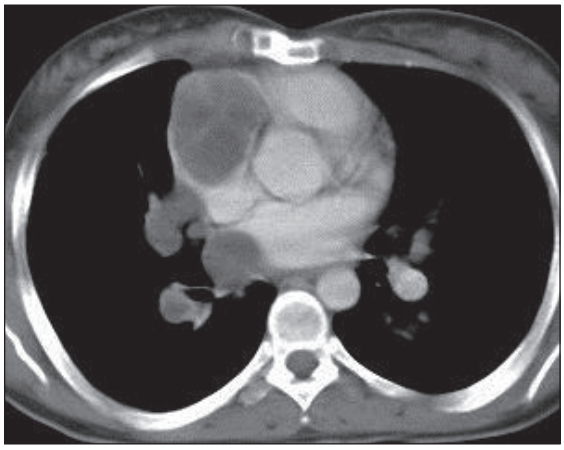

**Figure 5.** The tomographic imaging of CE on heart.

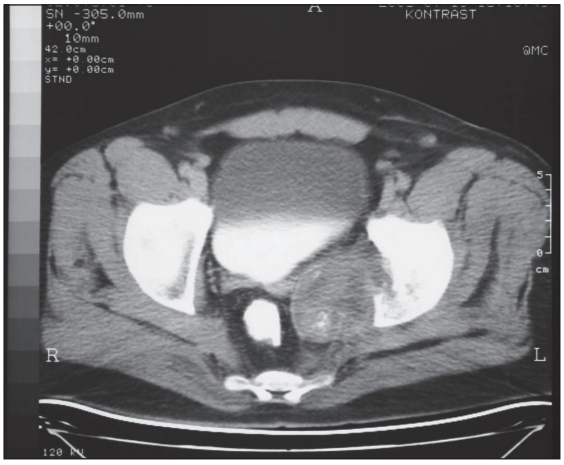


**Figure 6.** Osseos CE of left iliac spine.

*Kidney*

Eighteen patients with renal CE underwent surgery. In 17 cases (94%), CE lesions were isolated in the kidney, while in 1 case, concomitant liver lesions were detected. Among the 17 cases of isolated renal disease, 1 case had a history of a liver surgery for CE. Lesions were located in the right kidney (**Figure 4**) in 11 cases and in the left kidney in 7 cases. The IHA test was positive in 11 of 18 (61%) cases, while the WB test was positive in 7 of 7 cases, although 3 of these were IHA negative. During the follow-up period (median follow-up: 60 months), no evidence of recurrence was detected.

*Muscle and subcutaneous tissue (soft tissue)*

Fourteen patients with soft tissue CE underwent surgical treatment (**Figure 5**). The IHA test was diagnostic in 10 of 14 (71.4%) cases. Though WB test yielded positive results in 5 cases, the IHA test results were negative in 2 cases.

*Heart*

Thirteen patients with cardiac CE underwent surgical treatment. The first seven cases have been previously reported.[3] The most frequent symptoms were dyspnea (60%) and precordial pain (30%). In all cases except 1, echocardiography was performed as the first examination. After detecting CE lesions on echocardiography, radiological examinations (CXR, CT, and/or MRI) (**Figure 5**) and laboratory tests (IHA, WB) were performed in 12 of 13 patients. The IHA test results were positive in 11 of 13 (84.6%) cases. The WB test was performed in 6 cases, and positive results were obtained in all.

*Bone*

Fourteen patients with osseous CE underwent surgery in the Orthopedics and Neurosurgery clinics (**Figure 6**). The most common presentation was pain and swelling of the involved region. A pathologic fracture was detected in 2 patients. IHA tests were performed in 14 cases, and positive results were obtained in 12 cases. WB tests were performed in 6 cases, and a positive result was obtained in all cases. A CXR, an abdominal US, and/or a CT scan were performed in all cases to locate the systemic disease. In 2 cases, hepatic CE was established. In 10 cases, the disease was isolated in the bone. In the postsurgical period, recurrence was detected in the 18th month and in the 21st month in 2 patients in whom excision and curettage were performed. Hence, these patients had to undergo a second surgery. Unfortunately, the other 2 patients with long bone involvement, in whom reconstruction was performed

during the same surgery, experienced postsurgical infection and required a second surgery.

*Pancreas*

Three patients with pancreatic CE were treated surgically. In 2 cases, a preoperative diagnosis was successfully obtained by radiological examination, but in 1 case a laparotomy was required. The IHA test results were positive in both of these 2 cases. The WB test was performed only in 1 case, and a positive result was obtained. The third case was a 17-year-old male patient who was admitted with abdominal pain and jaundice. A US examination showed a pure CE lesion (5-cm mass) at the pancreatic head. CT and MR tests demonstrated a 5-cm-diameter CE mass around the head of the pancreas, and the ecstasy of both the choledochal and the pancreatic ducts indicated the possibility of a type I choledochal cyst. Magnetic resonance cholangiopancreatography showed that the cyst did not originate from the choledochal duct but displaced the choledochal duct laterally (**Figure 7**). Percutaneous transhepatic cholangiography showed lateral displacement of the choledochal duct, and the CE lesion did not fill with the contrast material. The serological test results (IHA) and the test for tumor markers were both negative. Histopathologic examination confirmed the diagnosis of a CE. No other organ involvement was detected on radiological examination.

*Ovary*

Two patients with ovarian CE were treated surgically. One was located on the right ovary and the other was located on the left ovary (**Figure 8**). The IHA and WB tests were positive in these 2 patients. One patient also had omentum involvement, which was misdiagnosed in another clinic as an ovarian malignancy. No evidence of recurrence was detected during the follow-up period (60 months).

*Thyroid*

One female patient had CE of the thyroid gland. She presented with a mass in the anterior aspect of the cervical area. A US examination revealed a 5  5 $cm^2$ cystic mass with characteristic features of CE in the right thyroid gland (**Figure 9**). A CT scan demonstrated an encapsulated cystic mass. An ultrasonographic examination of the liver and a chest x-ray were both normal. The IHA test results were positive. A CE lesion was completely removed with a right lobectomy.

*Breast*

A female patient with CE of the breast presented with a

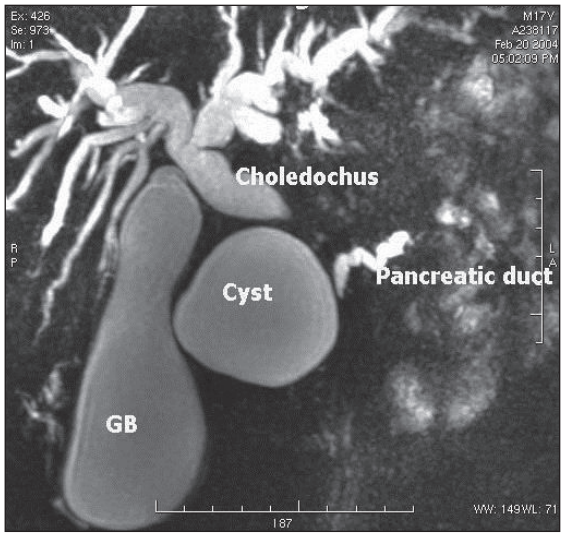


**Figure 7.** MRCP imaging of CE of pancreas.

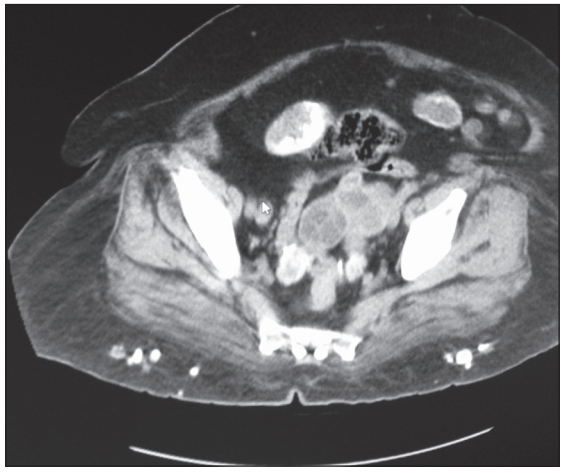

**Figure 8.** The tomographic image of bilateral adnexal CE.

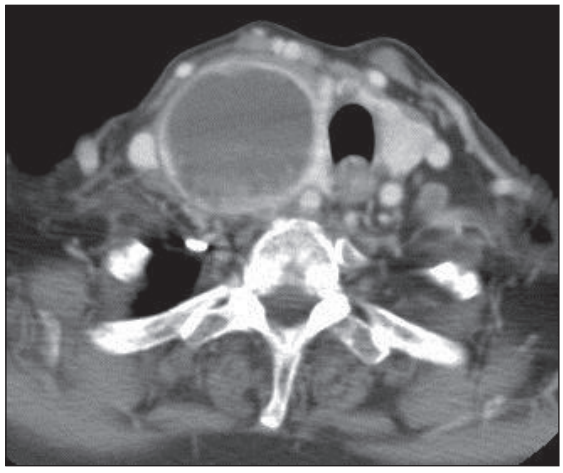

**Figure 9.** CE on the right neck originated from the right lobe of the thyroid.

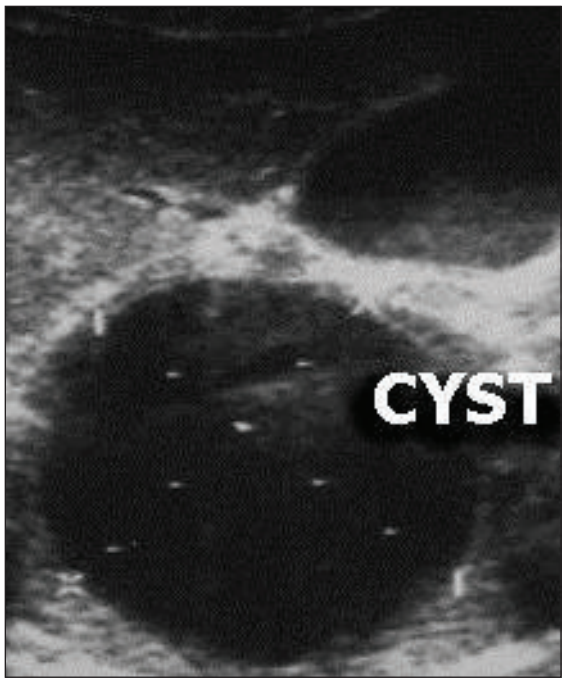


**Figure 10.** Ultrasonographic demonstration of breast CE.

painful mass on her right breast. On physical examination, a 5 5 $cm^2$ mobile mass with no auxiliary lymphadenopathy was detected. A US of the breast revealed a 5 4.5 $cm^2$ cystic lesion with daughter cysts circumscribed by means of a well-defined wall (**Figure 10**). An IHA test was performed for further work-up, and the result was positive.

## DISCUSSION

CE with unusual localizations may cause serious problems of diagnostic confusion. The combination of clinical history, radiological findings, and serological test results (especially the WB) are valuable in diagnosing extrahepatic CE. Immunodiagnosis can play an important complementary role. It is valuable not only for primary diagnosis but also for follow-up of patients after surgical treatment.[8-10,14] There are considerable differences in the specificity and sensitivity of the various tests. Ideally, a test should be specific with high sensitivity.[15] Detecting circulating *E granulosus* antigens in sera is less sensitive than detecting antibodies, which remains the method of choice. An IHA test has rather good sensitivity (between 60% and 100%) but poor specificity. WB allows molecular weight analysis of the antigens detected from the patient's serum with specific results.[15-17] WB serology is 80% to 100% sensitive and 88% to 96% specific for liver CE but less sensitive for lung (50%-56%) disease or other organ involvement (25%-26%).[15] Thirty-one (23.1%) of 134 extrahepatic CE cases were evaluated as negative with indirect hemagglutination (IHA). However, positive results were obtained in all 54 cases evaluated with a WB test including the 10 IHA-negative cases. It is suggested that this contrast with the published studies may be because of the decreased number of WB tests performed in extrahepatic and extrapulmonary cases in the previous published studies, or because 41 of our cases had a history of accompanying liver and/or lung lesions.

Diagnosis is easily established with US, CT, or MRI scans of patients who have characteristic findings such as calcification of the cyst wall, presence of daughter cysts, and detachment of the germinative membrane. However, CE with unusual sites of infection may cause serious problems in the differential diagnosis.[8-10,14,18,19] US is a noninvasive, accessible, inexpensive, highly sensitive (93%-98%), and specific (90%) imaging technique.[8,14,20,21] The advantages of US for the diagnosis and follow-up of subcutaneous and soft tissue CE are well recognized. Sonography reveals echo-free cysts, floating membranes, and daughter cysts. The radiological findings of CE involving skeletal muscles are similar to those involving parenchymal organs such as the liver, kidney, and spleen.[22] A multivesicular lesion is the characteristic of CE on US examination. The presence of small, dense, mobile echoes in the cyst is due to hydatid sand. The presence of small, dense mobile echoes in a cyst is specific or even pathognomonic of CE.[8,20-23] However, imaging methods are insufficient in the differential diagnosis of a simple type I CE cyst. Echocardiography is a valuable diagnostic method for cardiac CE.[3,24,25] In all but 1 case, the preoperative diagnosis was established by echocardiography during investigations for other suspected causes. The retrocardiac area or the posterior aspect of the heart can be better evaluated with TEE.[3,8] A CT and an MRI are further diagnostic evaluations.[3,8,26] A CT scan and/or an MRI should be performed in all suspected cases of CE or to elucidate the relationship of the cysts with adjacent organs and tissue for deciding the most appropriate surgical strategy. Further, these imaging techniques are essential for the preoperative evaluation of cases such as intracranial, spinal, or osseous CE.[8,19,26-31]

The peritoneal cavity and retroperitoneal CE accounted for 6% to 16% of cases in the published studies. The brain is also an uncommon site for CE. CNS involvement occurs in 2% to 3% of patients, and in endemic areas these CE lesions account for 2% of all intracranial space-occupying lesions.[8,27,32-37] Children are more frequently affected when compared with adults. It has been suggested that ductus arteriosus patency dur-

ing the neonatal period may be a cause of the increased vulnerability of children because it allows the passage of parasites from the periphery to the brain.[37-39] All CE were supratentorial and solitary, but no multiplicity was detected. Consistent with our series, cerebral CE reported in the published studies is usually supratentorial and solitary.[8,37-42]

Although it is rarely contaminated by Echinococcus (0.9%-8%), the spleen was one of the most frequently involved organ reported in the published studies.[9,10,38] In our study, spleen involvement was determined in 2.4% of all CE cases. Primary spleen involvement occurred in 10 (62.5%) cases. It is believed that the oncosphere may migrate to this location from the portal vein during bouts of increased abdominal pain.[10,12,39,43,44] CE of the urinary tract is uncommon, and the kidneys are the most frequently involved organs of the urinary tract. CE of the kidney occurs usually as isolated disease.[9,10,45-47] In our series, 94.4% of all renal CE occurred as isolated disease.

The involvement of the muscle and the soft tissue occurred in 2.1% of all CE cases and was observed in 10.5% of unusual CE cases. Echinococcosis of the musculoskeletal system is found in 0.5% to 4% of the patients suffering from CE ductus arteriosus. The involvement of muscle alone is much rarer. There are few reports of primary muscular CE, perhaps because the parasites have to cross the hepatic and pulmonary barriers to reach the muscles. Further, the presence of lactic acid in the muscles provides an inhospitable environment.[48,49] In contrast to the published studies, 9 isolated cases were found in our series.

The rate of cardiac involvement was 1.9% in our study and 0.2% to 2% in the published studies.[8,10,24] The myocardium of the left ventricle was reported to be the most frequently (75%) involved site.[10] In our study, 5 of 10 myocardium-localized cases and 1 pericardium-localized case were the result of secondary CE. Prousalidis and colleagues reported that the CE of the myocardium is considered a primary disease and that hydatid cysts of the pericardium are considered a secondary disease.[10] In fact, primary pericardial CE lesions are extremely rare.[24,26,50-53]

CE affects the bones in 1% of reported cases, and the pelvis or spine in 57% to 65% of cases.[22,54] While the pelvis and the spine were the most involved bones reported in the published studies, the rate of pelvic and spinal infection was 33% in our series,. We believe that the most important factor in the variation of rates compared to the published studies is the increased presence of intracranial intraosseous (parietal) CE in our series—these have been reported very rarely in the published studies. Usually, the diagnosis is established intraoperatively by an experienced surgeon. Although there have been advances in imaging techniques and improvements in surgical treatment combined with the use of antihelminthic administration, the published studies report a local recurrence and/or infectious complication rate of 50% in patients with CE of the bone.[22,54-57] In our series, recurrence occurred in 2 cases of cyst excision with curettage. Additionally, infection developed after simultaneous reconstruction of 2 other cases of long bone resected. Based on our experience, we recommend that prosthesis replacement should be performed in another sequence.

Pancreatic involvement has been reported in 0.25% to 0.75% of adult cases of CE, and the mode of infection is presumed to be hematogenous, However, local spread via the pancreatic or bile ducts has been suggested; peripancreatic lymphatic invasion has also been observed as a mode of infection.[9,10] Three cases of CE located in the pancreatic corpus or head (0.5%) were reported. The pre-operative diagnosis of CE of the pancreas may be difficult because it may be confused with a pseudocyst, choledochal cyst, simple cyst of the pancreas, cystic pancreatic neoplasm (such as cystadenocarcinoma), true congenital cyst, or post-traumatic pancreatic cyst. Radiological investigations and serological tests are valuable for the differential diagnosis; however, a diagnostic laparatomy could be necessary in some patients.[9,10,58,59] Radiological and serological tests were insufficient to diagnose CE in 1 case of our study, and diagnostic laparotomy was required.

The incidence of thyroid gland involvement was 0.15% in our series. CE presents as a solitary cystic lesion in the thyroid gland. The management of thyroid CE is surgical; complete excision should be the procedure of choice.[9,60-62] The breast is a rare primary site accounting for only 0.27% of all cases.[8,56] Although rare, CE should always be suspected in patients with palpable breast lumps, especially in endemic regions. The appearance of a mammary CE on US evaluation varies in all patients, and a variety of sonographic patterns have been previously described.[8,63-67] The presence of a CE lesion with a well-defined wall having racemose interior containing daughter cysts in a slowly growing breast mass is suggestive of a CE.[8,64,65] CE should be suspected while conducting the differential diagnosis of pelvic cystic masses.[68] As in our cases, primary ovarian CE may be confused with ovarian cystic tumors.

The recurrence rates of CE exhibited a large range based on the site of the disease (0%–50%).[17,22] In our study, recurrence occurred in 2 cases in which only

drainage was performed, and rupture occurred intraoperatively and involved the bone. In addition to these cases, recurrence occurred in retroperitoneum, which was treated with PAIR.

In conclusion, CE with unusual localizations may cause serious diagnostic confusion. The combination of the clinical history, radiological findings, and serological test results (especially the WB) are valuable for diagnosis. CE may develop in every organ of the human body and should be considered in the differential diagnosis of cystic masses, especially, in patients in regions where CE is endemic.

## REFERENCES


**1.** Akinoglu A, Bilgin I, Erkoçak EU. Surgical management of hydatid disease of the liver. Can J Surg 1985;28:171-4.
**2.** Demircan O, Baymus M, Seydaoglu G, Akinoglu A, Sakman G. Occult cystobiliary communication presenting as postoperative biliary leakage after Hydatid liver surgery: Are there significant preoperative clinical predictors? Can J Surg 2006;49(3):177-84.
**3.** Yaliniz H, Tokcan A, Salih OK, Ulus T. Surgical treatment of cardiac Hydatid Disease. Tex Heart Inst J 2006;33:333-9.
**4.** Sayek I, Tırnaksız MB, Do?an R. Cystic Hydatid disease: Current trends in diagnosis and management. Surg Today 2004;34:987-96.
**5.** Soylu L, Aydogan B, Kiroglu M, Kiroglu F, Javadzadeh A, Tuncer I, et al. Hydatic Cyst in the Head and Neck Area. Am J Otolaryngology 1995;16(2):123-25.
**6.** Togrul E, Kalacı A, Sarpel Y, Koltas IS, Ozbarlas S. What's your diagnosis? Diagnosis: Solitary Hydatid Cyst of the soleus muscle. Ann Saudi Med 2004;24(4):307-9.
**7.** Erman T, Tuna M, Gocer I, Ildan F, Zeren M, Cetinalp E. Intracranial intraosseous hydatid cyst. Neurosurg Focus 2001;11:1-3.
**8.** Engin G, Acunas B, Rozanes I, Acunas G. Hydatid disease with unusual localization. Eur Radiol 2000;10:1904-12.
**9.** Hamamcı EO, Besim H, Korkmaz A. Unusual locations of hydatid disease and surgical approach. ANZ J Surg 2004;74:356-60
**10.** Prousalidis J, Tzardinoglou K, Sgouradis L, Katsohis C, Aletras H. Uncommon sites of hydatid disease. World J Surg 1998;22:17-22.
**11.** Mushtaque M, Mir MF, Malik AA, Arif SH, Khanday SA, Dar RA. Atypical Localizations of Hydatid Disease: Experience from a single ınstitute. Niger J Surg 2012;18(1):2-7.
**12.** World Health Organization (2001) Echinococcosis in humans: clinical aspects, diagnosis and treatment. In: Eckert J, Gemmel MA, Melsin F-X, Pawlowski ZS (eds) WHO/OIE manual on echinococcosis in humans and animals: a public health problem of global concern. World Organization for Animal Health, Paris, pp 20–60
**13.** Eser I, Karabag H, Gunay S, Seker A, Cevik M, Ali Sak ZH, et al. Surgical approach for patients with unususlly located hydatid cysts. Ann Ital Chir 2013;18:84-6.
**14.** Balık AA, Celebi F, Basoglu M, Oren D, Yildirgan I, Atamanalp SS. Intra-abdominal extrahepatic echinococcosis. Surg Today 2001;31:881-4.
**15.** Biava MF, Dao A, Fortier B. Laboratory diagnosis of cystic hydatic disease. World J Surg 2001;25:10-14.
**16.** Aslan M, Yüksel P, Polat E, Cakan H, Ergin S, Öner YA, et al. The diagnostic value of Western blot method in patients with cystic echinococcosis. New Microbiol 2011;34(2):173-7.
**17.** Doiz O, Benito R, Sbihi Y, Osuna A, Clavel A, Gómez-Lus R. Western blot applied to the diagnosis and post-treatment monitoring of human hydatidosis. Diagn Microbiol Infect Dis 2001;41(3):39-42.
**18.** Orhan Z, Kara H, Tuzuner T, Sencan I, Alper M. Primary subcutaneous cyst hydatic disease in proximal thigh: an unusual localisation: a case report. BMC Musculoskeletal Disorders 2003;4;1-4.
**19.** Demirtas M, Usal A, San M, Birand A. Hydatid disease presenting as cardiac tamponade. A case history. Angiology 1996;47:601-7.
**20.** WHO Informal Working Group. International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings. Acta Tropica 2003;85:253-61.
**21.** Gharbi HA, Hassine W, Brauner MW, Dupuch K. Ultrasound examination of the hydatid liver. Radiology 1981;139:459-63.
**22.** Arazi M, Erikoglu M, Odey K, Memik R, Ozdemir M. Primary echinococcus infestation of the bone and muscles. Clin Orthop 2005;432:234-41.
**23.** Izol V, Eken A, Aridogan IA, Koltas S, Tansug Z. Acute urinary retention due to cystic echinococcosis: A case report. Can Urol Assoc J 2012;6(5):192-4.
**24.** Ibn Elhadj Z, Boukhris M, Kammoun I, Halima AB, Addad F, Kachboura S. Cardiac hydatid cyst revealed by ventricular tachycardia. J Saudi Heart Assoc 2014;26(1):47-50.
**25.** Kanadasi M, Demirtas M, San M, Ozer C, Soyupak SK, Kısacıkoglu B. Mobile right atrial hydatid cyst with multiorgan involvement. Catheter Cardiovasc Interv 2000;49:204-7.
**26.** Inzirillo F, Giorgetta C, Ravalli E. Pericardial echinococcosis: Unusual presentation in a non-endemic region. Asian Cardiovasc Thorac Ann 2014;22(9):1126-7.
**27.** Altınors N, Bavbek M, Caner HH, Erdogan B. Central nervous system hydatidosis in Turkey: a cooperative study and literature survey analysis of 458 cases. J Neurosurg 2000;93:1-8.
**28.** Vizcaychipi KA, Sosa S, Camicia F, Santillán G, Casalins M, Nigro Mdel C. Retroperitoneal hydatidosis secondary to hepatic hydatid cyst. Rev Argent Microbiol 2012;44(4):278-82.
**29.** Sachar S, Goyal S, Goyal S, Sangwan S. Uncommon locations and presentations of hydatid cyst. Ann Med Health Sci Res 2014;4(3):447-52.
**30.** Aksungur EH, Oguz M, Bicakci K, Altay M. Cerebral hydatid lesion with daughter cysts: MR and CT findings. Am J Radiol 1994;163:1269-70.
**31.** Comert RB, Aydingoz U, Ucaner A, Arikan M. Water-lily sign on MR. Imaging of primary intramuscular hydatidosis of sartorius muscle. Skeletal Radiol 2003;32:420-3.
**32.** Kapila K, Verma K. Sapiration cytology diagnosis of echinococcosis. Diagn Cytopathol 1990;6:301-3.
**33.** Bagga PK, Bhargava SK, Aggarwal N, Chander Y. Primary subcutaneous inguinal hydatid cyst: diagnosis by fine needle aspiration cytology. J Clin Diagn Res 2014;8(8):11-3.
**34.** Bagatur AE, Ugur F, Zorer G. Primary giant hydatid cyst in the thigh. Acta Orthop Traumatol Turc 2002;36:72-75.
**35.** Das S, Kalyani R, Kumar U, Kumar HM. A varied presentation of hydatid cyst: a report of four cases with review of literature. Indian J Pathol Microbiol 2007;50(3):550-2.
**36.** Sing P, Mushtad D, Verma n, Mahajan NC. Pelvic hydatidosis mimicking a malignant ovarian tumor. Korean J Parasitol 2010;4883:263-5.
**37.** Bozkurt M, Bozkurt DK, Cil AS, Karaman M. Primary ovarian and pararectal cysts mimicking pelvic endometriosis. Acta Med Iran 2012;50(12):839-42.
**38.** Ciurea, AV, Fountas KN, Coman TC, Machinis TG, Kapsalaki EZ, Fezoulidis NI, et al. Long term surgical outcome in patients with intracranial hydatid cyst. Acta Neurochir 2006;148:421-6.
**39.** Kulacoglu IH, Oruc MT, Kocaerkek Z. Unusual Localization of hydatid disease: an evaluation of 77 cases. Turk J Gastroenterol 2001;12(4):299-302.
**40.** Lunardi P, Missori P, Lorenzo ND, Fortuna A. Cerebral hydatidosis in childhood: a retrospective survey with emphasis on long term follow up. Neurosurg 1991;29:515-8.
**41.** Ersahin Y, Mutluer S, Guzelbag E. Intracranial hydatid cyst in children. Neurosurg 1993;33:219-25.
**42.** Madeo J, Zheng X, Ahmed S, Ramos De Oleo R. Primary cerebral echinococcosis presenting as long-standing generalized weakness. Germs 2013;3(2):63-6.
**43.** Schott A, Husak R, Kempf M. Splenic cysts as an incidental finding - what's next? Dtsch Med Wochenschr 2014;139(42):2132-5.
**44.** Sawarappa R, Kanoi A, Gupta M, Pai A, Khadri SI. Isolated splenic hydatidosis. J Clin Diagn Res 2014;8(6):3-4.
**45.** Yılmaz Y, Kosem M, Ceylan K. Köseoglu B, Yalçinkaya I, Arslan H, et al. Our experience in eight8 cases with urinary hydatid disease: A series of 372 cases held in nine different clinics. Int J Uro 2006;13:1162-67.
**46.** Angulo JC, Sanchez CM, Diego A, Escribano J, Tamayo JC, Martin L. Renal echinococcosis: clinical study of 34 cases. J Urol 1997;157:787–94.
**47.** Goel ML, Agarwal MR, Misra A. Percutaneous drainage of renal hydatid cyst: early results and follow-up. Br J Uro 1995;75:724–8.
**48.** Burgazli KM, Ozdemir CS, Beken Ozdemir E, Mericliler M, Polat ZP. Unusual localization of a primary hydatid cyst: a subcutaneous mass in the paraumbilical region. Eur Rev Med Pharmacol Sci 2013;17(13):1766-8.
**49.** Garcia-Alvarez F, Torcal J, Salinas JC, Navarro A, García-Alvarez I, Navarro-Zorraquino M, et al. Musculoskeletal hydatid disease: A report of 13 cases. Acta Orthop Scand 2002;73:227–31.
**50.** Narin N, Mese T, Unal N, Pinarli S, Cangar S. Pericardial hydatid cyst with a fatal course. Acta Pediatr Jpn 1996;38:61-2.
**51.** Cakir O, Eren N, Kılınc N. Cardiac hydatic cyst causing cerebral emboli in a child. Pediatr Cardiol 2002;23:555-6.
**52.** Byard RW, Bourne AJ. Cardiac echinococcosis with fatal intracerebral embolism. Arch Dis Child 1991;66:155-6.

**53.** Kumar Paswan A, Prakash S, Dubey RK. Cardiac tamponade by hydatid pericardial cyst: a rare case report. Anesth Pain Med 2013;18;4(1):9137.
**54.** Markasis P, Markaki S, Prevedorou D, Bouropoulou V. Echinococcosis of bone: Clinicolaboratory findings and differential diagnostic problems. Arch Anat Cytol Pathol 1990;38:92–94.
**55.** Ebrahimi A, Assadi M, Saghari M, Eftekhari M, Gholami A, Ghasemikhah R, Assadi S. Whole body bone scintigraphy in osseous hydatosis: a case report. J Med Case Rep 2007;19;1:93.
**56.** Nourbakhsh M, Shemshaki H, Zarezadeh A, Etemadifar MR, Mazoochian F. Recurrent Hydatosis at the Site of Non-union Humerus Fracture. Int J Prev Med 2012;3(9):660-3.
**57.** Yıldız Y, Bayrakcı K, Altay M, Saglik Y. The use of polymethylmethacrylate in the management of hydatid disease of bone. J Bone Joint Surg 2001;83:1005–8.
**58.** Choudhary NS, Kumar N, Puri R. An unusual cause of a pancreatic space-occupying lesion. Gastroenterology 2014;146(3):12-3.
**59.** D'Alia C, Lo Schiavo MG, Tonante A, Gagliano E, Taranto F, Bonanno L, et al. Pancreatic echinococcosis. Report of a clinical case and review of the literature. Chir Ital 2003;55:581-4.
**60.** Oksuz S, Pektas E, Yavuz M, Aksungur N, Cayir Y, Akcay MN. An unusual cause of hoarseness: hydatid cyst of the thyroid. Trop Biomed 2013; 30(4):642-4.
**61.** Ahmad S, Jalil S, Saleem Y, Suleman BA, Chughtai N. Hydatid cysts at unusual sites: Reports of two cases in the neck and breast. J Pak Med Assoc 2010;60(3):232-4.
**62.** Hmidi M, Touiheme N, Rbai M, Messary A. Isolated Hydatid cyst of the neck: an unusual site. Eur Ann Otorhinolaryngol Head Neck Dis 2012;129(2):108-10.
**63.** Vega A, Ortega E, Cavada A, Garijo F. Hydatid cyst of the breast: mammographic findings. AJR Am J Roentgenol 1994;162:825-6.
**64.** Taori KB, Mahajan SM, Hirawe SR. Hydatid Disease of Breast. Ind J Radiol Imag 2004;14:57-60.
**65.** Afroz N, Chaurasia JK, Maheshwari V, Singh G. Hydatid cyst: unusual presentation as 'breast lump'. BMJ Case Rep 2014;12; 2014.
**66.** Geramizadeh B, Boob R, Talei AR, Rasekhi A. Fine needle aspiration cytology in hydatid cyst of the breast. Acta Cytol 2003;47:701-2.
**67.** Elverici E, Barça AN, Erhuner Z, Sayın B. Hydatid cyst: An unusual solid breast mass. Clin Imaging 2013;37(3):577-9.
**68.** Agarwal S, Shah A, Kadhi SK, Rooney RJ. Hydatid bone disease of the pelvis. A report of two cases and review of the literature. Clin Orthop Relat Dis 1992;280:251–5.