Hydatid Cyst Disease Manifesting with Extensive Abdominal and Pelvic Involvement: Case Report

Yaygın Abdominal ve Pelvik Tutulumla Ortaya Çıkan Hidatik Kist Hastalığı

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ABSTRACT Hydatid cyst disease is a zoonotic parasitic infection and has worldwide distribution. Although the disease primarly affects the liver, it can be seen in almost any anatomic region depending on its hematogenous dissemination. In this case report, we presented a 13 year-old female patient who was complaining of severe abdominal pain and imaging studies revealed extensive hydatid cyst disease. She had hydatid cysts in the liver and also in the abdominal and pelvic cavities, probably due to dissemination of the disease from the liver. We described ultrasonography, computed tomography and magnetic resonance imaging findings of the disease. Indirect hemagglutination test for Echinococcus granulosus confirmed our diagnosis and the patient was placed on albendazole treatment. Hydatid cyst disease should be kept in mind whenever a cystic lesion is detected in any anatomic location in the body.

Key Words: Tomography scanners, X-Ray computed; magnetic resonance imaging; ultrasonography; echinococcosis; parasites

ÖZET Hidatik kist hastalığı zoonotik bir paraziter enfeksiyon olup dünyada yaygın bir dağılım gösterir. Hastalık primer olarak karaciğeri etkilediği halde, hematojen yayılımına bağlı olarak neredeyse vücutta her anatomik bölgede görülebilir. Olgu bildirimizde şiddetli karın ağrısı şikayeti olan 13 yaşında bir kız hastayı sunduk ve yapılan görüntüleme çalışmaları yaygın kist hidatik hastalığını ortaya çıkardı. Hasta karaciğerindeki kist hidatiklerin dışında muhtemelen karaciğerden hastalığın yayılımından dolayı abdominal ve pelvik kavite içerisinde de kistlere sahipti. Makalede hastalığın ultrasonografi, bilgisayarlı tomografi ve manyetik rezonans görüntüleme bulgularını tanımladık. Echinococcus granulosus için yapılan indirekt hemaglütinasyon testi tanımızı doğruladı ve hastaya albendazol tedavisine başlandı. Vücutta herhangi bir anatomik lokalizasyonda kistik bir lezyon ile karşılaşıldığı zaman, hidatik kist hastalığı olabileceği de akılda tutulmalıdır.

Anahtar Kelimeler: Bilgisayarlı tomografi; manyetik rezonans görüntüleme; ultrasonografi; ekinokokkozis; parazitler

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Hydatid cyst disease (HCD) is a parasitic infection caused by *Echinococcus granulosus* and it is endemic in many regions of the world. Although it mainly affects the liver and the lung, due to its hematogenous dissemination, it can also occur anywhere in the body. Radiological findings demonstrate a wide range appearance from being a purely cystic lesion to a completely solid mass. Hydatid disease can present as a solitary lesion or may be multiple. Calcification is more common in the hydatid disease of the liver, spleen and kidney.[1] Imaging methods consisting of ultrasonography (US), computed tomography (CT) and magnetic reso-

nance imaging (MRI) are useful for delineating the disease. HCD is also endemic in the south-east region of the Turkey[2] and in this case report we presented a child demonstrating hydatid disease with multiple site involvement in the abdomen.

CASE REPORT

A 13 year-old female patient was admitted to our hospital complaining of abdominal pain. In the emergency department, her physical examination was found normal and she was given analgesics for pain. But following a short-term relief, her abdominal pain became more severe and therefore she was referred to our US department for abdominal evaluation. Her routine blood and urine test results were within normal limits. Abdominal US examination (Aplio, SSA-770 ; Toshiba, Tokyo, Japan) revealed multiple hypoechoic cystic masses with varying sizes in the liver, abdominal peritoneal cavity and pelvic region (Figure 1). They all demonstrated a purely cystic appearance and therefore we interpreted them as type 1 HCD except for the lesion which was located at segment 5 in the liver. This lesion had septations and membranous structures within, so it was classified as a type 2 lesion. Following US, we performed an abdominal CT examination (Somatom Sensation 16 ; Siemens, Erlangen, Germany) for further characterization of these cystic appearance masses. On CT, multiple hypodense masses were detected throughout the liver, abdominal cavity and pelvic region and none of these lesions demonstrated calcification which is regarded as an important finding for HCD (Figure 2). Similar to US, all of the lesions were purely hypodense and only the segment 5 liver lesion had a heterogenous appearance due to its membranous and septational structures. None of the lesions demonstrated contrast enhancement, except the segment 5 liver lesion that showed septational contrast enhancement. For better delineating the extent of the disease, we carried out an abdominal MRI by a 1.5 tesla superconducting magnet. (GE, Signa, Milwaukee, Wisconsın, USA) This examination also revealed multiple cystic masses that extensively involved the abdomen and the pelvis, showing hypointense signal intensity on T1 weighted images and hyperintense signal intensity on T 2 weighted images without having any contrast enhancement (Figure 3).

For diagnosis, we considered HCD with a primary location in the liver from where it showed a hematogenous dissemination to the abdominal and pelvic cavities. Following our consideration of HCD, a chest x-ray was primarly carried out for ruling out lung involvement which was found normal. Then an indirect hemagglutination test was performed and resulted positive for *Echinococcus granulosus* (1/972). The patient was consulted with the pediatric surgery department and they did not recommend surgery or percutaneous drainage due to disseminated disease. Therefore medical treatment was initiated and the patient was placed on albendazole treatment (15 mg / kg / day x 2). The medical treatment was planned for 6 months. Fol-

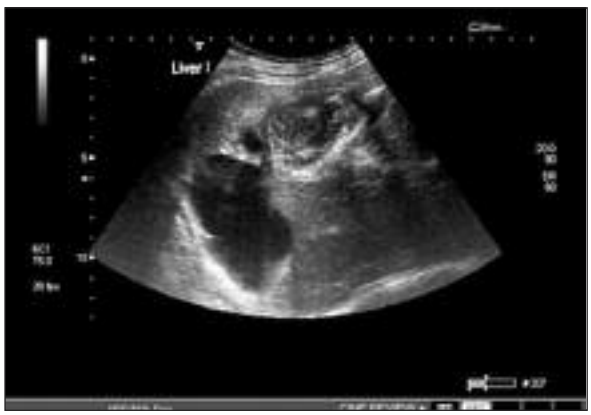

FIGURE 1: Ultrasonography image showing type 1 and type 2 exophytic hydatid cysts in the liver parenchyma.

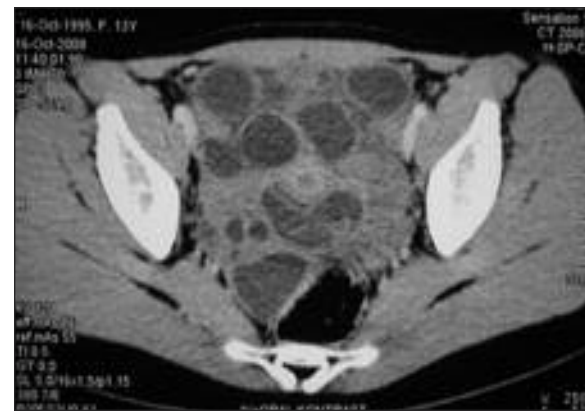

FIGURE 2: Axial CT image demonstrates multiple hypodense cystic masses occupying the pelvic region.

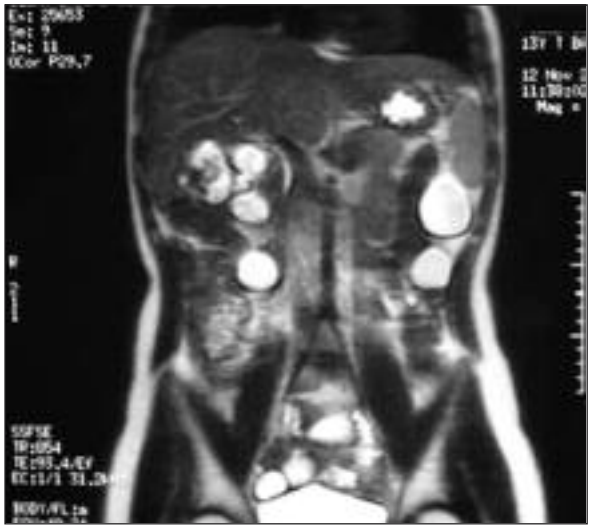

FIGURE 3: Coronal fat-suppressed T 2 weighted image revealing multiple hyperintense cystic lesions throughout the liver and the peritoneal cavity consistent with extensive hydatid cyst disease.

lowing 3 months treatment, the patient was evaluated and although she was clinically doing well, there was no regression neither in her indirect hemagglutination test level nor in her cystic lesions regarding the number and size. She is closely being followed up by both the pediatrics and radiology clinics and will be controlled at the end of the 6 months medical treatment.

DISCUSSION

HCD is a worldwide zoonosis caused by two types of *Echinococcus.* Where *Echinococcus granulosus* is the more common and *Echinococcus multilocularis* is the less common but more invasive type, the latter may sometimes even mimic malignancy. Although the classical type hydatid disease is well depicted, findings related to its complications and the disease occuring in unexpected anatomic regions are less frequently described in the literature.[3]

The liver is the most common location of HCD and the right lobe is the most frequently involved portion of the liver. Arslan A. et al[4] have shown that in 43 out of 55 cases (%78.18) of their series, the right lobe involvement were found. Hydatid cysts in the liver may be solitary or multiple and imaging findings can vary depending on the stage of the cyst growth. A solitary type 1 hydatid cyst may be sometimes difficult to distinguish from a simple epithelial cyst on the basis of imaging findings. Simple cysts do not demonstrate internal structures but in type 1 hydatid cyst, multiple echogenic foci within the lesion due to hydatid sand can be determined when the patient changes the position. Multiple hydatid cysts can exhibit different stages. Type 2 disease may appear as a well-defined fluid collection with a localized split in the wall and floating membranes inside the cavity. In our case report, we demonstrated both type 1 and type 2 hydatid cysts occuring simultaneously in the liver. Multivesicular hydatid cysts may manifest as well-defined fluid collections in a honeycomb pattern with multiple septa representing the walls of the daughter cysts.

US is proven to be the most sensitive modality for the detection of membranes, septae, and hydatid sand within the cyst. Calcification is seen on radiography in 20-30% of hydatid cysts and usually manifests with a curvilinear or ringlike pattern representing calcification of the pericyst. During the natural evolution toward healing, dense calcification of all components of the cyst occurs. This can indicate the death of the cyst.[5] CT and conventional radiography are the best modalities for detecting calcification. CT has a high sensitivity and specificity for hepatic hydatid disease. Intravenous administration of contrast material is not necessary unless complications are suspected, especially infection and communication with the biliary tree. Although US is the initial diagnostic tool for HCD, CT is more useful in terms of revealing calcification and detecting daughter cysts and therefore is regarded to be more sensitive and accurate than US in the differential diagnosis.[6] Hepatic hydatid cysts may have a low signal intensity rim on T 2 weighted MR images. This finding has been proposed to be a characteristic sign of HCD. It probably represents the collagen-rich outer layer of the hydatid cyst and is generated by the host.[7] This finding can also be observed in our case. When present, daughter cysts are seen as cystic structures attached to the germinal layer that are hypointense relative to the intracystic fluid on T 1 weighted images and hyperintense on T 2 weighted images.[7]

Hydatid cysts may use the natural routes provided by the liver capsule, ligaments and peritoneum to progress beyond the boundaries of the liver. The two most common routes of exophytic growth are the bare area of the liver and the gastrohepatic ligament. Peritoneal echinococcosis is almost always secondary to hepatic disease, although a few unusual cases of primary peritoneal involvement has been described.[8] The overall frequency of peritoneal disease in cases of abdominal hydatid disease is approximately 13 %. Although most of these cases are related to previous surgery for hepatic disease, in 12 % of cases, asymptomatic microruptures of hepatic cysts into the peritoneal cavity may occur,[8] as seen in our case where a previous hepatic hydatid cyst surgery does not exist. These cysts are generally multiple and can arise anywhere in the peritoneal cavity. They can be undetectable until they become large enough to produce symptoms. Peritoneal hydatid disease may grow and occupy the entire peritoneal cavity, simulating a multiloculated mass. This pathological condition has been referred to as encysted peritoneal hydatidosis.

HCD can be treated by medical, surgical and percutaneous modalities. Albendazole is used for the medical treatment of hydatid disease and is found to be superior to mebendazole. Medical treatment indications are inoperability, recurrence and extrahepatic disease involvement.[9] The usual dose of orally-administered albendazole is 10-15 mg/kg/day in two divided doses. Treatment is typically administered as 1-6 monthly cycles separated by 10-14 day intervals. US-guided cyst puncture is another choice of treatment which has been used successfully in selected patients, although anaphylactic and allergic reactions due to spillage of the cyst contents have occured. Surgical therapy in HCD is indicated for large cysts with multiple daughter cysts, superficially located single liver cysts which have a risk of rupture, complicated cysts such as those accompanied by infection, compression or obstruction, and cysts located in vital organs . But surgical therapy may cause to morbidity, mortality or recurrence in some cases. Therefore medical therapy can be selected in uncomplicated cysts and in patients who have high risks for surgery.[10]

In conclusion, although HCD primarly affects the liver and has typical imaging findings, secondary involvement due to hematogenous dissemination can occur and the hydatid cysts can reach any anatomic location in the body. Therefore when a cystic lesion is encountered in any other anatomic location, HCD should always be remembered in the differential diagnosis.

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