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Multiple Cerebral Hydatic Cyst Developed After Operation of Cardiac Hydatic Cyst: A Case Report

Kardiyak Hidatik Kist Operasyonu Sonrası Gelişen Multiple Serebral Kist Hidatik: Olgu Sunumu

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ABSTRACT

Hydatid cyst is a zoonotic disease and is an important health problem, especially in developing countries. Hydatic cysts are typically observed in the liver and lungs. Cardiac and brain involvement are rare manifestations. Cardiac hydatic cysts are usually located in the left ventricle. Brain involvement is frequently seen as a primary cerebral cyst and is almost always solitary. However, secondary intracerebral cysts are also seen as a result of cardiac cysts rupturing into the left ventricle spontaneously or iatrogenically, and these are usually multiple. Herein, we report a case that has two rare clinical manifestations of hydatid cysts.
Keywords: Echinococcosis, hydatid cyst, zoonoses

ÖZ

Zoonotik bir hastalık olan kist hidatik, özellikle gelişmekte olan ülkelerde önemli bir sağlık sorunudur. Kist hidatik tipik olarak karaciğer ve akciğerlerde görülmektedir. Kardiyak ve beyin tutulumu hastalığın nadir görülen tutulumlarıdır. Kardiyak kistler genellikle sol ventrikülde görülmektedir. Beyin tutulumu ise sıklıkla primer serebral kist olarak görülür ve çoğunlukla soliterdir. Ancak kardiyak kistlerin spontan veya iyatrojenik olarak sol ventriküle rüptüre olması sonucu sekonder intraserebral kistler de görülmektedir ve bunlar genellikle multipldir. Bu yazıda, kist hidatiğin iki nadir klinik tutulumunun birlikte görüldüğü bir olgu sunulmuştur.
Anahtar Kelimeler: Ekinokokkoz, kist hidatik, zoonozlar

INTRODUCTION

Hydatic cyst (HC) is a zoonotic infection caused by *Echinococcus granulosus* (*E. granulosus*) (1). The disease is widely endemic in regions where livestock farming is prevalent and is included in the list of notifiable diseases in Türkiye (2). The parasite has a life cycle that involves two main hosts. The definitive host is usually dogs, while humans act as intermediate hosts and become infected through fecal-oral ingestion of *E. granulosus* eggs. These eggs develop into oncospheres in the intestine and migrate through the systemic circulation to various organs. Haematogenously spreading oncospheres often cause cysts in the liver, followed by cysts in the lung, and spleen, and very rarely in the brain, heart, and musculoskeletal system (3). Here, we report a case of multiple cerebral HCs that had been previously undergone surgery for intracardiac HCs.

CASE REPORT

An 8-year-old male patient applied to the outpatient clinic with complaints of fainting, vomiting, and chest pain. Physical examination findings were normal, but electrocardiography showed ST elevation in the right leads, and the troponin level was elevated. He was consulted to the cardiology department with a

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prediagnosis of myocarditis. Transthoracic echocardiography revealed a mass lesion in the interventricular septum. Mediastinal magnetic resonance imaging (MRI) showed a 37x25x45 mm septal cystic mass without significant contrast enhancement in the interventricular septum (Figure 1). On contrast-enhanced thoracoabdominal computed tomography (CT) revealed a 48x46x58 mm echinococcosis stage 1 (CE1) cystic lesion (according to the world health organization classification of cyst) in liver segment 7 and isodense mass lesion without calcification (Figure 2). Brain CT imaging was normal at the time. Albendazole 15 mg/kg/day (2 doses daily) was started preoperatively. A pathological examination of the interventricular mass operation revealed a HC.

Three years after the initation presentation, he presented with headache, vomiting, and occasional blurred vision for 20 days. Vital signs and laboratory tests were normal. On neurological examination, sensory, and motor functions and reflexes were normal. On brain CT imaging, there were multiple smoothly circumscribed cystic lesions, the largest of which was 46x49x48 mm in size, without compression effect and edema, without calcification, and located in the supratentorial region. There was no contrast uptake on contrast-enhanced brain CT (Figure 3). Brain MRI showed multiple well-circumscribed supratentorial cystic lesions that were hypointense on T1-weighted images, hyperintense on T2-weighted images, and without perilesional edema on fluid-attenuated inversion recovery images (Figure 4). The patient preferred a tertiary hospital for surgical treatment. Albendazole was started before surgery. Cysts in the brain were removed by craniotomy surgery. The patient had no complaints at the first postoperative visit and physical examination was normal. Postoperative brain imaging is shown in Figure 5.

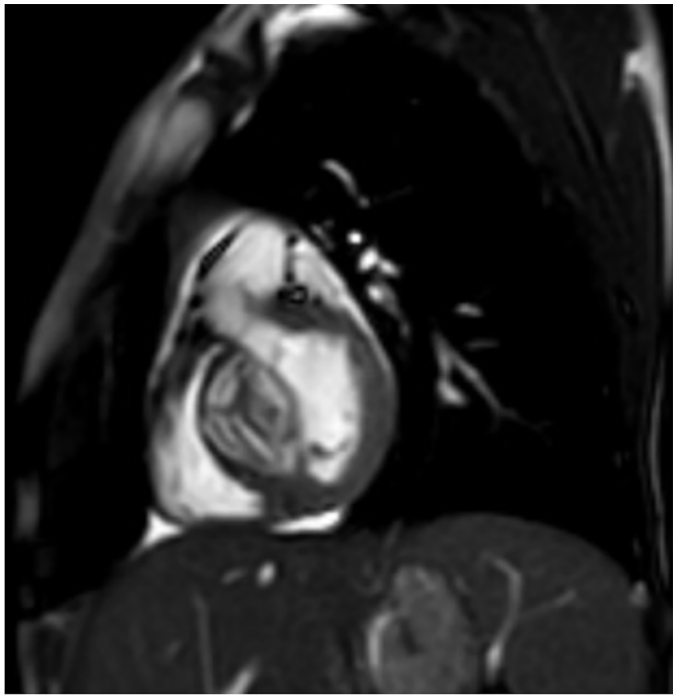

Figure 1. Mediastinal MRI showed a 37x25x45 mm septal cystic mass without significant contrast enhancement in the interventricular septum
MRI: Magnetic resonance imaging

DISCUSSION

HCs may involve all organs, primarily the liver and secondarily the lungs. Cardiac HC is very rare and occurs in 0.5-2% of all HCs. Cysts are usually found in the left ventricular myometrium (4). The symptoms of a cardiac HC vary according to its location. The cyst may be asymptomatic due to its slow growth, or it may cause valvular regurgitation, atrioventricular defect, arrhythmias, or

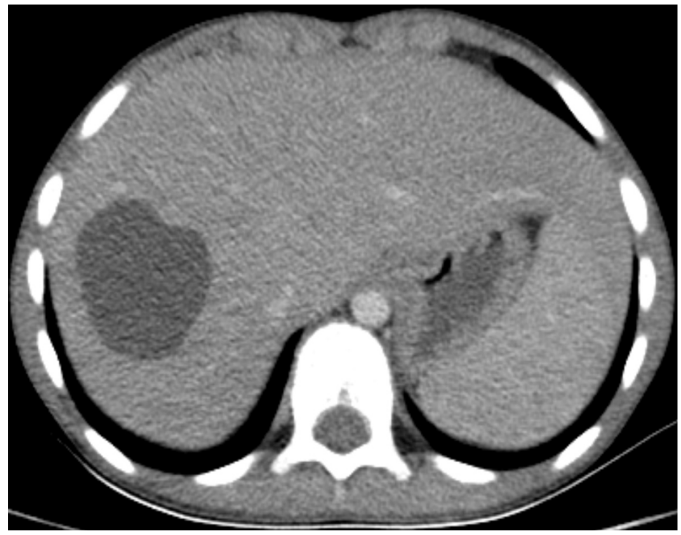

Figure 2. The contrast-enhanced thoracoabdominal CT revealed a 48x46x58 mm cystic lesion in liver segment 7
CT: Computed tomography

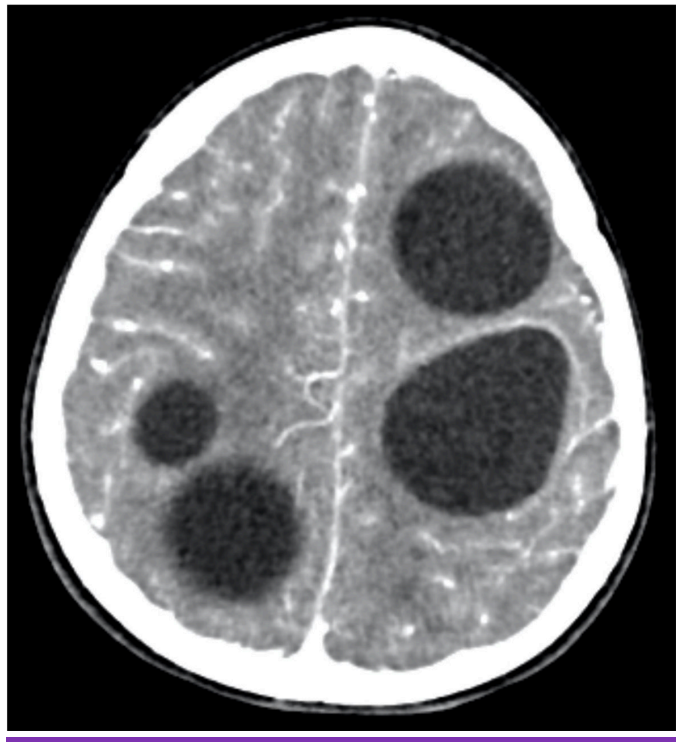

Figure 3. Brain CT showed multiple smoothly circumscribed cystic lesions, the largest of which was 46x49x48 mm in size, without compression effect and edema, without calcification, and located in the supratentorial region
CT: Computed tomography

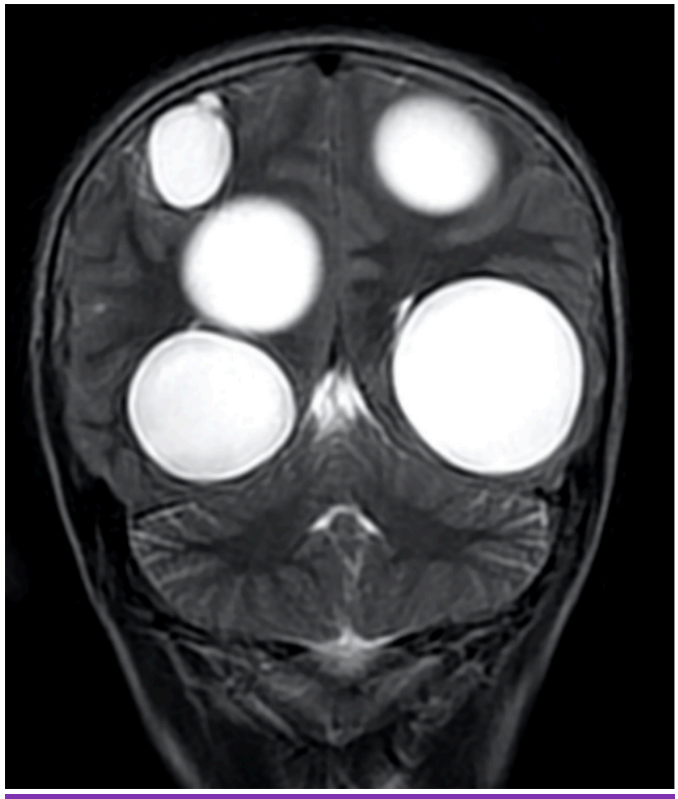

Figure 4. Brain MRI showed multiple well-circumscribed supratentorial cystic lesions that were hypointense on T1-weighted images, hyperintense on T2-weighted images, and without perilesional edema on FLAIR images

MRI: Magnetic resonance imaging, FLAIR: Fluid-attenuated inversion recovery

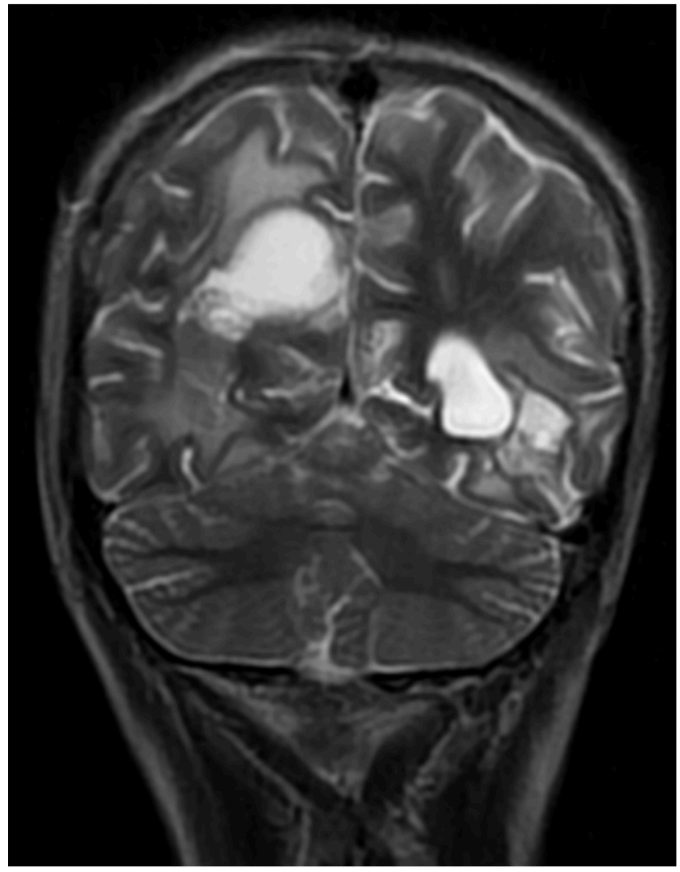

Figure 5. Postoperative brain MRI showed an increase in intensity consistent with edema and around the postoperative cavities

MRI: Magnetic resonance imaging

rupture of the cyst leading to peripheral and pulmonary embolism, pericardial effusion, and sudden death due to anaphylaxis (5). In our case, the cyst was located in the interventricular septum and caused symptoms of fainting, vomiting, and chest pain. Brain involvement is seen in only 1-2% of HC cases. 80% of patients with cerebral HCs are in the pediatric age group (6). Although the clinical presentation may be asymptomatic depending on the size and localization of the cyst, it may lead to increased intracranial pressure or focal neurological symptoms such as headache, vomiting, visual blurring or loss, hemiparesis, seizures and behavioral changes. Cerebral HCs can grow 1-10 cm per year (7). While primary intracranial cysts are usually solitary, secondary cysts are multiple. Secondary cerebral HC cases constitute more than 50% of cerebral hydatid cyst patients. The cysts are supratentorial in the middle cerebral artery region (8). In our case, it was located supratentorial and caused symptoms of headache, vomiting, and occasional blurred vision. Brain CT was normal at initial presentation and the presence of multiple cysts suggested that the cardiac HC might have multiple dissemination to the cerebral parenchyma as a complication of the operation.

HCs are usually diagnosed by clinical findings, serological and imaging methods. In some cases, serological tests may be false negative, and imaging methods may give better results. In addition, diagnosis, staging, and follow-up can be easily made with radiological imaging. In radiological imaging, MRI is very functional to evaluate the nature of the cyst. Localization of the internal and external structural features of the cyst can be easily evaluated. HCs may be unilocular or multilocular and may have daughter vesicles, membrane separation, and wall calcification. The diagnosis can be easily made in these cases, but another differential diagnosis should be considered when solidifying hydatid cysts.

Radiological evaluation of the cerebral HC and the hypointense peripheral ring is a typical finding on MRI T2A images (6). Perilesional edema or wall calcifications are usually not seen around them. Perilesional edema and contrast enhancement usually indicate rupture or are complicated by secondary infection. Rupture of the cyst may cause recurrent cysts in the subarachnoid space, intracranial and spinal regions (9). Arachnoid cysts, porencephalic cysts, pyogenic abscesses, neurocysticercosis, and cystic tumors should be considered in the differential diagnosis of cerebral HC (10). Treatment of cardiac and cerebral HC is medical and surgical. Preoperative albendazole is used as a supportive treatment to reduce postoperative recurrences (11). In our case, we used albendazole before cardiac and brain surgery.

CONCLUSION

In conclusion, this case is important in terms of its contribution to the literature, as it presents a rare occurence of HCs. If it comes to mind in the diagnosis, the diagnosis can be easily made with radiological imaging findings. It should be kept in mind in the differential diagnosis, particularly for individuals residing in endemic areas.

* Ethics

Informed Consent: A consent form was completed by all participants.

* Authorship Contributions

Surgical and Medical Practices: Ö.R.K., Concept: İ.M., Ö.R.K., Y.Ç., Design: İ.M., Y.Ç., Data Collection or Processing: Ö.R.K., Y.Ç., Analysis or Interpretation: İ.M., Y.Ç., Literature Search: İ.M., Ö.R.K., Writing: İ.M., Y.Ç.

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