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CASE REPORT

Isolated Giant Splenic Hydatid Cyst Treated with Spleen-Sparing Surgery

Dalak Koruyucu Cerrahi ile Tedavi Edilen İzole Dev Dalak Hidatik Kisti

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Abstract

Isolated splenic hydatid cyst is a very rare parasitic infection. This case report presents the diagnosis and treatment process of giant splenic hydatid cyst treated with open spleen-sparing surgery. A 27-year-old female patient presented to the general surgery clinic with abdominal complaints. On computed tomography scan, she had a mass, 150 × 130 × 100 mm in size, at the medial side of the spleen. Spleen-sparing surgery was applied to the patient, who seemed to have a splenic hydatid cyst in the preoperative examinations. The patient was discharged without complications on the fourth postoperative day. Postoperative pathology was compatible with hydatid cyst. Albendazole treatment continued for two more months postoperatively.

Keywords: Albendazole; Hydatid cyst; Spleen

The causative agent of hydatid cyst (HC), popularly known as cyst disease, is a parasite called Echinococcus granulosus. The main source of this parasite is meat-eating animals such as dogs, wolves, and foxes. HC is rarely visible in all organs and soft tissues such as the liver, spleen, kidney, and brain. The most frequently affected organ by HC is the liver. On the other hand, spleen involvement is a rare condition with a rate of 2.5–5.8%, even in endemic areas.[1] HC is especially common in the Eastern Anatolia of Turkey.

HCs of the spleen may present with abdominal pain, abdominal mass, or compression symptoms.[2] Splenomegaly and palpable mass may be detectable on physical examination in case of large cysts. They are also asymptomatic and can be detected incidentally by imaging studies such as ultrasonography (USG) or computed tomography (CT). Diagnosis becomes clear only with serological tests and imaging tools. Although USG and CT findings are not specific for HC, they are the most valuable examination tools for diagnosis. Wall calcification can be seen in advanced cysts at plain X-ray radiography.[3] Patients diagnosed with splenic HC should undergo systemic screening for possible involvement in other organs.

The main treatment is the surgical evacuation of the cyst/removal of the cyst or splenectomy in cases that are impossible and occupy most of the spleen volume. These surgical procedures can be performed with open surgery and laparoscopic surgery. Although it is not a gold standard op-

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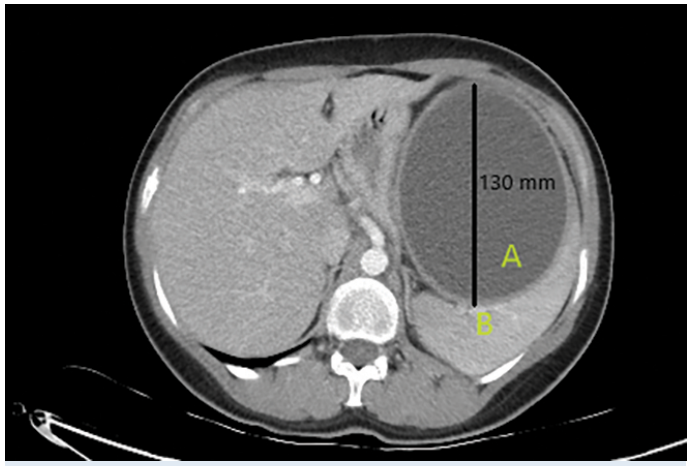

Figure 1. Axial image of the splenic hydatid cyst (A: Cyst, B: Spleen).

tion for surgery, the risk of cyst rupture and contamination is higher during laparoscopy. Open surgery is preferable in terms of ease of exploration, especially in large cysts.

This case report presents the diagnosis and treatment process of a giant splenic HC treated with opens spleen-sparing surgery.

Case Report

A 27-year-old female patient presented to our general surgery department in July 2021. She had occasional nausea, abdominal cramping attacks, vomiting, and gastroesophageal reflux-like symptoms such as heartburn and food regurgitation for about six months. She was offered surgery before and did not accept surgery due to the COVID-19 pandemic. Her complaints had increased significantly in the last two months. She had no history of additional disease or operation in her history.

In her vital signs evaluation, her blood pressure was 124/68 mm Hg, her heart rate was 82 beats/minute, her oxygen saturation (room air) was 98%, and her body temperature was 36.9°C. The patient was 72 kilograms in weight. She had abdominal pain in deep palpation and a palpable mass in the left upper quadrant on abdominal physical examination. Other system examinations, including digital rectal examination, were normal.

There was no pathology at basic hematological parameters and biochemical parameters. Her Echinococcus indirect hemagglutination antigen (IHA) test was positive in 1/640 titer. On USG, there was a mass with a diameter of 120 mm at the medial side of the spleen. On CT scan, there was a cystic mass approximately 150 × 130 × 120 mm in size at the medial side of the spleen (Fig. 1–3). There is no additional cyst on the CT scan.

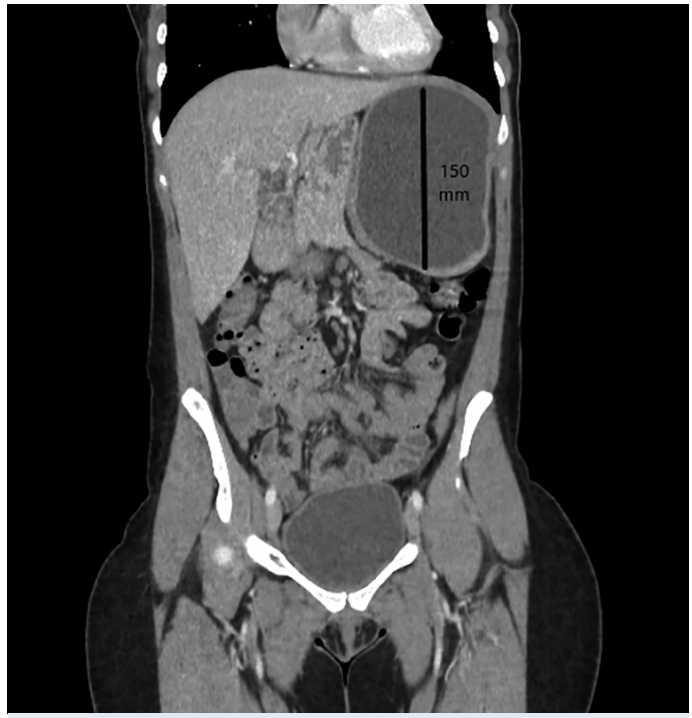

Figure 2. Coronal image of the splenic hydatid cyst.

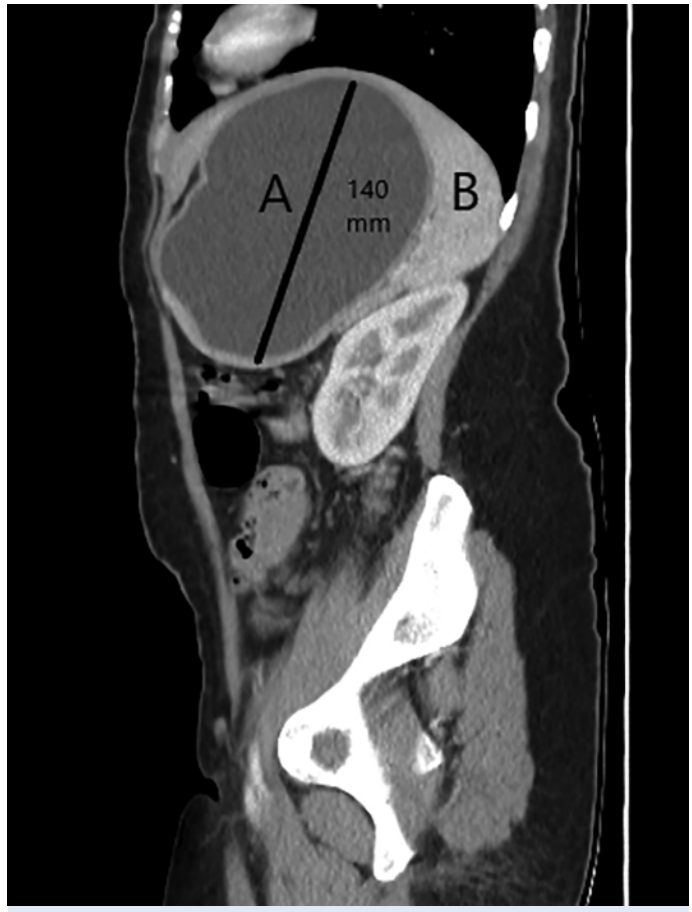

Figure 3. Sagittal image of the splenic hydatid cyst (A: Cyst, B: Spleen).

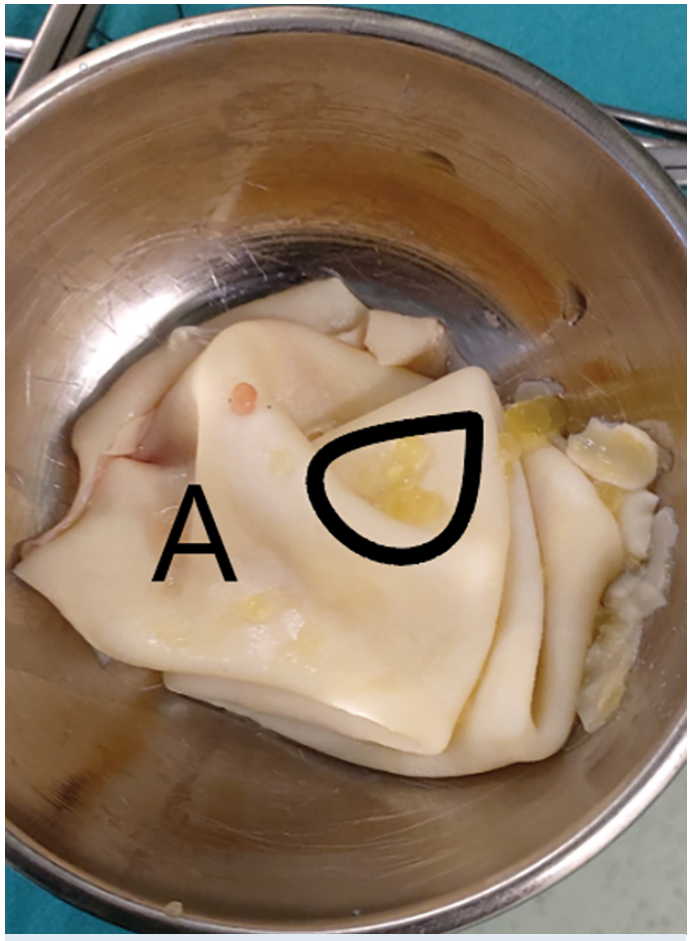

Figure 4. Intraoperative image of the hydatid cyst (A: Germinative membrane, daughter vesicles in the black circle).

Splenic HC was considered, and albendazole treatment (400 milligrams every 12 hours) was initiated for 3 weeks. After a 7-day break, open surgery was performed with a midline incision. At the intraoperative evaluation, a cystic mass was seen medial of the spleen. Sponges impregnated with the hypertonic solution were placed around the mass. The hypertonic solution was infused into the cyst cavity and waited for 15 minutes. The spleen capsule was opened from the lower part of the spleen, and the cyst cavity was observed. There were multiple daughter cysts with large germinative membranes (Fig. 4, 5). Deroofing with external drainage was performed. The operation was terminated by placing a drain in the cyst cavity.

The patient was followed in the service after surgery. Oral feeding was opened in the early postoperative period. On postoperative day 1, about 150 cc of serohaemorrhagic fluid came from the drain. On the 2nd day and the 3rd day after surgery, about 50 cc of serous fluid came from the drain. After drain removal on postoperative day 4, she was discharged without complications. Postoperative pathology was compatible with HC. Albendazole treatment continued for two more months. There was no fluid collection or cyst recurrence in the operation area at the 3rd month postoperative USG control.

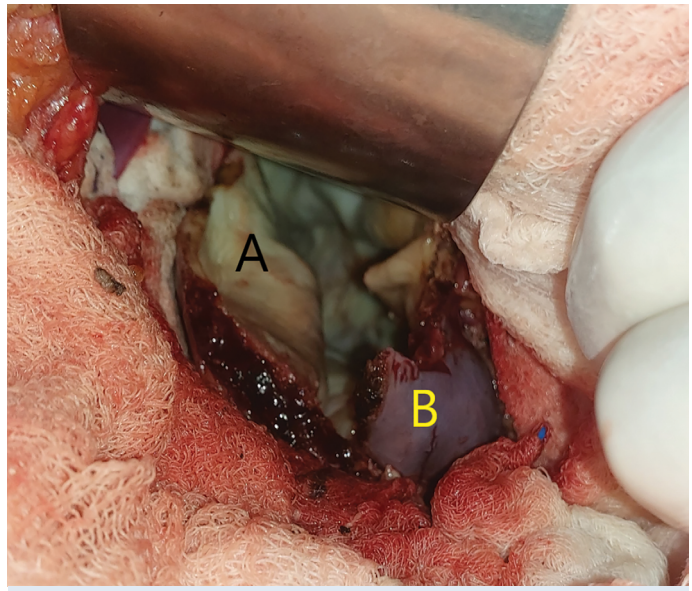

Figure 5. Intraoperative image of the hydatid cyst (A: Cyst cavity, B: Splenic tissue).

Discussion

HC is a disease that has been seen worldwide since ancient times. Echinococcus granulosus causes HC in humans. HC is a zoonotic disease that causes significant public health problems and serious economic losses in our country. HC is especially common in Turkey's Eastern Anatolia and Southeastern Anatolia regions.[4] The disease can be seen in all tissues and organs of the body, although it occurs in the liver at 75% and in the lungs at 15%. Although the spleen is the third most frequently involved organ, spleen involvement is a rare condition even in endemic areas. Berlot described the first splenic HC in an autopsy study in 1790. The prevalence of splenic hydatid can reach up to 8% in various studies.[5] However, an isolated splenic HC is a rare condition, as in our case.

The clinical presentations of the HC vary depending on organ involvement and cyst size. The cysts at the lungs grow faster, but the cysts at the liver, spleen, and kidneys grow slower.[6] In the liver localization, symptoms such as pain, nausea, vomiting, and sometimes itching and jaundice occur. Pulmonary involvement may cause respiratory distress, cough, bleeding from the mouth, and chest pain. In intracranial involvement, headache, vomiting, loss of consciousness can be seen. Cardiac involvement may include cardiac arrhythmias, signs of infarction, and even rupture of the heart wall.

While small or calcified hydatic cysts may be asymptomatic, large hydatic cysts may exert pressure or may rupture. The cysts may grow 1–5 cm in size per year or stay silent for years. Splenic cysts compress the stomach and present with left upper quadrant pain, heartburn, early satiety, nausea, vomiting, and weight loss. In addition, splenic HC may apply with ileus-like symptoms by pressing on the transverse colon.[7] In our case, the patient had left upper quadrant pain, nausea, vomiting, weight loss, and gastroesophageal reflux-like symptoms such as heartburn and regurgitation.

Basic hematological and biochemical tests, IHA tests, and radiological screening tools help diagnose splenic HC. Eosinophilia and higher immunoglobulin E serum levels can be seen, but they are not specific. ELISA and IHA are commonly used diagnostic tests. The sensitivity of IHA is 60–100%, but its specificity is reportedly low.[2] IHA remains positive even up to 1 year after eradication of the organism. It is very difficult to diagnose splenic HC with conventional radiography alone. However, if the cyst is severely calcified, it may be suspected on plain X-ray radiography. In addition, the left diaphragm is displaced upwards, the stomach is displaced to the right, and the splenic flexure is displaced downwards. USG and CT show the locations of surrounding tissues and organs more effectively. CT gives more accurate information than USG. It shows the size and location of the cyst better. However, a definitive diagnosis can be made with these two imaging tools in almost all cases. In addition, female cyst and hydatid sand, which are findings specific to echinococcal infection, can be identified in these examinations. Uncalcified cysts without daughter vesicles cannot be differentiated from other benign cysts by CT and USG. In addition, USG is also used in postoperative follow-up. In our case, no pathology was detected in the basic laboratory tests, but her IHA test was positive. In addition, both USG and CT were used in the diagnosis process. However, only USG was used in the follow-up.

HC's management options include surgery, percutaneous drainage, drug therapy, and observation stage. Although treatment arrangements are made according to HC diagnostic classification of the World Health Organization, there is no definitive treatment algorithm. The number of septa in the cyst and cyst size are important criteria in the treatment. Surgery is recommended in cases such as single cyst diameter >100 mm, superficial cyst with risk of rupture, ruptured cyst, cysts with secondary infection or bleeding, cysts compressing vital structures, and cysts with multiple daughter vesicles.[8] The surgical options used to manage splenic HC >50 mm are mainly divided into splenectomy and spleen-sparing procedures. Spleen-sparing procedures have gained importance as death rates from sepsis-associated with total splenectomy are 4% and 1.9% in children and adults, respectively. Studies have shown no significant difference in disease recurrence or complications when total splenectomy or spleen-sparing procedures are compared.[9] However, studies show that spleen-sparing surgery has better results in younger patients.[10]

The main aim of splenic HC surgery is to clear the cyst and eliminate the residual cavity. This aim can be provided with open surgery and laparoscopic surgery. Laparoscopic surgery is still controversial because the risk of spillage due to pneumoperitoneum increases during laparoscopy.[11] Although there is no consensus on the choice of surgery, antiparasitic treatment should be used in the preoperative and postoperative periods. World Health Organization suggests that albendazole (or mebendazole) therapy should be initiated 4–30 days before surgery and continued for at least 1 month after surgery.[12] However, the ideal duration of antiparasitic treatment is uncertain. Since our case was young and suitable for spleen-sparing surgery, splenectomy was not performed on the patient. In addition, the patient was given antiparasitic treatment both in the preoperative period (for 3 weeks) and in the postoperative period (for 8 weeks).

Conclusion

Splenic HC is a rare parasitic health problem. HC can be asymptomatic or cause compression symptoms when it reaches large sizes. Although there is not a definitive method for diagnosis, the combination of laboratory, and imaging tools is helpful in diagnosis. The main treatment is surgery combined with antiparasitic therapy. Surgery can be performed open or laparoscopically, depending on the cyst size and location, and the surgeon's experience. In order to reduce the complication and sepsis rate, splenectomy should be avoided as much as possible, and spleen-sparing surgeries should be applied.

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Informed Consent: Written informed consent was obtained from patients who participated in this study.

Authorship Contributions: Concept: TK, TA; Design: TK, TA; Supervision: TK, TA; Materials: TK, TA; Data Collection or Processing: TK, TA; Analysis or Interpretation: TK, TA; Literature Search: TK, TA; Writing: TK, TA; Critical Review: TK, TA.

Conflict of Interest: None declared.

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