Case Report: Primary Giant Splenic Echinococcal Cyst Treated by Laparoscopy

Maira A. Arce, Himerón Limaylla, Maria Valcarcel, Hector H. Garcia, and Saul J. Santivañez*

Instituto Peruano de Parasitología Clínica y Experimental (INPPACE), Lima, Perú; Department of Surgery, Hospital Nacional Dos de Mayo, Lima, Perú; Department of Microbiology, School of Sciences, Universidad Peruana Cayetano Heredia, Lima, Perú; Center for Global Health, Tumbes, Universidad Peruana Cayetano Heredia, Lima, Perú; Cysticercosis Unit, Instituto Nacional de Ciencias Neurológicas, Lima, Perú

Abstract. Cystic echinococcosis (CE) is a zoonosis caused by the larval stage of the dog tapeworm *Echinococcus granulosus*. Liver and lungs are the most commonly affected organs whereas splenic infection is rarer and its primary involvement occurs in less than 2% of abdominal CE. We report a case of primary giant splenic hydatid cyst in a 75-year-old Peruvian woman that was laparoscopically removed without any complications, perioperative prophylactic chemotherapy with albendazole 400 mg twice a day 5 days before, and 7 days after the surgical procedure was administered, postoperative recovery was uneventful, and; at her 3-month follow-up the patient remains asymptomatic and an abdominal computed tomography scan demonstrated a cystic cavity of 15 cm diameter with no daughter vesicles, neither other abdominal organ involvement. This case is in line with the existing literature on laparoscopic treatment of splenic cystic hydatid disease, suggesting that laparoscopic treatment is a safe and effective approach for large splenic hydatid cysts to be preferred to open surgical techniques.

INTRODUCTION

Cystic echinococcosis (CE) is a zoonosis caused by the larval stage of the dog tapeworm *Echinococcus granulosus*, and is endemic in livestock-raising areas around the world.^{1,2} Typically, the cycle of *E. granulosus* is maintained when dogs eat infected sheep viscera containing the larval stage of the parasite.³ Humans in close contact with dogs infected with the adult tapeworm stage of the parasite are at risk for infection, as they become accidental intermediary hosts. In endemic areas, as much as 3–9% of the general population may harbor liver or lung echinococcal cysts.^{4–6}

Humans are usually infected during childhood, and develop symptoms after several years from infection. Symptoms are related to enlargement or complication of the cyst(s) (i.e., rupture or infection).³ The organs most commonly involved are the liver (70%) followed by the lungs (25%), while other organs (e.g., brain, bones, spleen, kidneys) are more rarely involved, accounting for approximately 5% of cases.¹

In clinical case series, splenic involvement is reported in 0.5–8% of CE cases.⁷ Primary involvement of spleen is rarer and occurs in less than 2% of abdominal CE.⁸ The diagnosis of splenic CE is based on imaging findings and confirmed by a positive serological result. The suggested treatment of large cysts is surgical extraction either by a preserving technique (cystectomy and lobectomy) or more radically by splenectomy.^{9–11} Recent reports describe the use of laparoscopic techniques in the treatment of large splenic cystic hydatid disease (CHD) cases.^{12–14} We report here a patient with a giant primary echinococcal cyst of the spleen treated by laparoscopy and a review of the literature on laparoscopic treatment of splenic CHD.

CASE PRESENTATION

A 75-year-old Peruvian woman with a history of 2 months of abdominal pain in the left hypochondrium was admitted to the General Surgery Department at the Hospital Nacional

Dos de Mayo, Lima, Peru. The patient came from Apurímac, in the southern Peruvian highlands, where she lived until the age of 19 years when she moved to a coastal city. The patient referred frequent contact with livestock and dogs in her native town. One month before admission, the abdominal pain increased in intensity and frequency, later accompanied by intermittent nausea and vomiting. Two weeks before admission, frequent episodes of dizziness appeared and affected her daily activities.

Preoperative imaging, hematological and biochemical evaluations were performed. A plain chest X-ray showed an elevation of left diaphragm until the seventh intercostal space that was apparently produced by an intra-abdominal mass, but no cysts were seen (Figure 1A). The patient reported to have undergone an abdominal ultrasound, but no documentation was available in her clinical record. Abdominal computed tomography (CT) scan showed a 16.5 × 13 cm cystic lesion in the upper pole of spleen with a partially calcified wall that contained four daughter vesicles, and compressed and displaced surrounded viscera (stomach, tail of pancreas, and left kidney); no other intra-abdominal cysts were observed (Figure 1B and C). Blood tests revealed low hemoglobin (11.2 g/dL), mild leukopenia (2,910 cell/mm³), and eosinophilia (5%). Renal and liver function tests were in the normal range. Finally, an enzyme-linked immunoelectrotransfer blot assay for CE demonstrated specific antibody bands of 8, 16, and 21 kDa.¹⁵

Hand-assisted laparoscopic surgery was then performed: pneumoperitoneum using carbon dioxide (12 mm Hg) was produced by a Veress needle inserted in the supraumbilical area; one 10-mm working port was inserted by the Hasson technique and two 5-mm ports were placed, one just below to the left of xiphoid cartilage for positioning and the second parallel to the umbilicus in the left midclavicular line for aspiration. After locating the spleen cyst, gauzes impregnated with hypertonic saline were positioned around the incision site; a 3 × 3 cm incision was made and initial aspiration of cyst contents was performed. After this step, five cycles of injection and aspiration of hypertonic saline as scolicedal agent were performed and then all the cyst contents were aspirated, leaving a large open cavity with calcified areas (Figure 1D). A tubular drain was introduced through the midclavicular line port and positioned lateral to the lower pole of the spleen.

*Address correspondence to Saul J. Santivañez, Instituto Peruano de Parasitología Clínica y Experimental, Ca. Hera 391, Ate, Lima, Perú. E-mail: ssantiva@jhsph.edu

FIGURE 1. (A) Plain chest X-ray showing an elevation of left diaphragm until the seventh intercostal space. (B) Computed tomography scan of abdomen showing a large splenic hydatid cyst with a calcified wall (arrow), (C) containing daughter vesicles (arrow). (D) Opened cavity left after all the cyst content was aspirated showing calcified areas (arrow).

FIGURE 2. (A) Abdominal ultrasound at 3-month follow-up visit showing a cavity (marked by circle) with a heterogeneous content (anechoic and hypoechoic). (B) A computed tomography scan performed at the same time showing a residual cystic cavity of 15 cm diameter with calcified wall (arrow) and without daughter vesicles.

TABLE 1
Clinical data of patients with splenic cystic hydatid disease treated by laparoscopy

Author	Country/year	Sex	Age	Other organs involved	Cyst's size (cm)	Cyst type	Localization	Surgery done	Surgical complications	Treatment pre-surgery (time)	Treatment post-surgery (time)	Recurrence (follow-up time)
Ballaux and others ²²	Belgium/1997	M	26	NS	5 × 5	Partially calcified with daughter vesicles	Central segments	Splenectomy	No	NS	NS	NS
Gharaibeh ¹⁴	Jordan/2001	M	45	No	8 × 12	Unilocular cyst	Lower polar segment	Partial cystectomy	No	Albendazole 400 mg c/ 12 hours (NS)	Albendazole 400 mg c/ 12 hours (2 months)	None (13 months)
Georgescu and others ¹³	Romania/2004	M	69	Yes*	11 × 11	NS	NS	Partial cystectomy	No	Albendazole 10 mg/kg/day (10 days)	Albendazole 10 mg/kg/day (3 months)	NS
Fuente and others ²³	Mexico/2007	M	69	NS	8 × 7 × 6	Calcified cyst	Upper polar and central segment	Splenectomy	No	NS	NS	NS
Diaconescu and others ¹²	Romania/2008	M	44	NS	12.5 × 12.5	Unilocular cyst	Central segments	Splenectomy	No	NS	Albendazole 20 mL c/ 24 hours (3 months)	None (24 months)
Polat and others ²⁵	Turkey/2009	F	38	NS	6 × 7	Cyst with daughter vesicles	Upper polar segment	Partial cystectomy	No	Albendazole 400 mg/day (28 days)	NS	None (8 years)
Mishra and others ²⁴	India/2010	M	6	Yes†	5.4 × 5 × 3.5	Cyst with daughter vesicles	Upper polar segment	Partial cystectomy	No	Albendazole 10 mg/kg/day (NS)	Albendazole 10 mg/kg/day (3 months)	None (18 months)
Akkoyun and others ²¹	Turkey/2011	F	10	No	7 × 6 × 6	Cyst with daughter vesicles	Lower polar segment	Partial cystectomy	No	Albendazole 10 mg/kg/day (NS)	Albendazole 10 mg/kg/day (NS)	None (21 months)
Singal and others ²⁶	India/2011	M	14	No	9 × 7	Cyst with daughter vesicles	Hilar region in the upper polar segment	Splenectomy	NS	NS	NS	NS
Arikanoglu and others ¹¹	Turkey/2012	F	32	No	6 × 5	NS	Peripheral of the spleen	Partial cystectomy	Hemorrhage, intra-abdominal abscess	NS	NS	NS
Vezakis and others ²⁷	Greece/2012	M	44	No	NS	Calcified cyst	NS	Splenectomy	No	NS	Albendazole 10 mg/kg/day (3 months)	None (12 months)
Arce and others	Peru/2012	F	75	No	15 × 15	Cyst with daughter vesicles and calcified wall	Upper polar segment	Partial cystectomy	No	Albendazole 400 mg c/ 12 hours (5 days)	Albendazole 400 mg c/ 12 hours (3 months)	None (3 months)

F = female; M = male; NS = nonspecified.
 * History of liver, lung, and intra-abdominal involvement.
 † History of lung involvement.

Postoperative recovery was uneventful and the drain was removed at day 7 after surgery, and the patient was discharged 3 days later. Perioperative prophylactic chemotherapy with albendazole at 400 mg twice a day was indicated from 5 days before surgery to 40 days after. The patient stopped treatment at day 7 for personal reasons. At her 3-month follow-up visit, the patient remains asymptomatic and her abdominal ultrasound demonstrates a cavity with a heterogeneous content (anechoic and hypoechoic) of 12 cm diameter (Figure 2A); additionally, a CT scan performed at the same time demonstrated a cystic cavity with no daughter vesicles of 15 cm diameter, with no other intra-abdominal organ neither peritoneal involvement (Figure 2B).

DISCUSSION

Clinical series of CE report spleen involvement in 0.5–8% of all cases, usually associated with the presence of cysts in the liver or another intra-abdominal organ.^{7,16} In around 2% of cases, the spleen is the only organ involved (primary infection).⁸ Peru is highly endemic for CE, with rates of up to 9% in the highland.^{4–6} Although the growth rate of echinococcal cysts is extremely variable, it basically depends on the organ compliance, reaching larger sizes in the lungs and growing at slower pace in the liver, spleen, or kidneys.¹⁷ If a cyst does not become complicated (infected or ruptured), symptoms related to the enlargement of the cyst only appear several years after the infection. Not infrequently, the diagnosis is incidentally established when a patient is evaluated for other diseases.¹⁸ This 75-year-old woman presented with symptoms related to the presence of a large abdominal mass, and she has been asymptomatic until 2 months before. An additional symptom, dizziness, was likely related to a supine hypotensive syndrome caused by the intra-abdominal mass.¹⁹ Her town of residence was located in a region considered not endemic for CHD, so it is likely that she contracted the infection decades ago when living in the highlands. We cannot rule out entirely a more recent infection, since migration patterns and transport of animals can establish foci of infection in apparently nonendemic cities.²⁰

To date, there are only 11 other cases of laparoscopically treated spleen cysts reported (Table 1).^{11–14,21–27} In these cases, the diagnosis of splenic CHD was based on imaging findings, complemented by positive serology in only three of them.^{13,14,25} In this particular case, the initial finding was an elevated left hemi diaphragm on chest X-rays (suggesting a large intra-abdominal mass), and then the splenic cysts was well visualized on abdominal CT scan. Typical findings included a partially calcified and four daughter cysts. Although the baseline ultrasound report was not available, the CT findings suggest that the cyst would have corresponded to a Stage 3B of the World Health Organization ultrasound classification.²⁸ Positive serology confirmed the diagnosis in this case, although negative serology is frequent in uncomplicated cysts.^{29,30}

Traditionally, surgery is the recommended treatment option for large splenic cysts. Radical techniques (i.e., splenectomy) are associated with sepsis-related mortality in 4% and 1.9% of treated children and adults, respectively,^{10,31} and were thus replaced by more conservative procedures, either partial splenectomy, or cystectomy.^{11,32} More recently, other techniques as percutaneous drainage of the cyst by injection and

reaspiration of a scolical agent (ethanol or polidocanol) and laparoscopy have been proposed as good alternatives.^{33,34} The first case treated by laparoscopy was reported in 1997 and thereafter another nine cases have been reported, including four with total splenectomy and six with partial cystectomy, all in cysts larger than 5 cm diameter (Table 1). No recurrences or seeding with new lesions were seen in the five cases followed for more than a year (follow-up time 13 months–8 years). Reflecting the situation in liver CE, perioperative administration of albendazole was done in some of the cases, with large variations in time of treatment. In our case, after 3 months of treatment, the patient is asymptomatic, neither postoperative complications nor recurrence were observed, but recurrence and evidence of spillage would usually be observed in longer periods.^{12,21,24,25}

Despite being an uncommon presentation, splenic CE should be considered in patients with a splenic cyst, who had lived in an endemic area, and the diagnosis is well established using ultrasound or abdominal CT, except for the CE1 type that may be difficult to differentiate from non-parasitic cystic lesions. Our case and the existing literature suggest that laparoscopic treatment is a safe and effective approach for large splenic echinococcal cysts and it can replace open surgical techniques.

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Authors' addresses: Maira A. Arce and Saul J. Santivañez, Instituto Peruano de Parasitología Clínica y Experimental (INPPACE), Lima, Perú, E-mails: mairaarce@hotmail.com and ssantiva@jhsph.edu. Himerón Limaylla and Maria Valcarcel, Division of Surgery, Hospital Nacional Dos de Mayo, Lima, Perú, E-mails: mari_valcarcel@hotmail.com and hlimaylla@hotmail.com. Hector H. Garcia, Department of Microbiology, School of Sciences, Universidad Peruana Cayetano Heredia, Lima, Perú, and Cysticercosis Unit, Instituto Nacional de Ciencias Neurológicas, Lima, Perú, E-mail: hgarcia@jhsph.edu.

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