



Primary Retroperitoneal Hydatid Disease Causing Secondary Hypertension: A Case Report and Review of the Literature

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ABSTRACT

Hydatid disease is caused by the parasite *Echinococcus granulosus*. Humans are accidental intermediate hosts, and the parasite commonly affects the liver and the lungs. Primary retroperitoneal hydatid disease is extremely rare. We present a rare case of a primary retroperitoneal hydatid cyst with secondary hypertension treated through surgical means.

INTRODUCTION

Hydatid disease is a zoonosis produced worldwide by *Echinococcus* tapeworm larvae. The most commonly involved organs are the liver (75%) and the lungs (15%), with only 10% of cases occurring in other parts of the body [1]. Retroperitoneal involvement was always thought to be secondary to a rupture of the liver or spillage during liver surgery for hydatid disease [2]. Primary retroperitoneal hydatid disease is extremely rare, accounting for only 0.8% of cases [2,3]. We present a rare case of a primary retroperitoneal hydatid cyst with secondary hypertension that was treated with surgery.

CASE REPORT

A 65-year-old female presented with dull aching pain on the left side of her abdomen for a duration of 6 months. She was recently diagnosed with hypertension 3 months prior to presentation. Urinary symptoms were absent. On examination, her blood pressure was 160/100 mm Hg. There was a cystic, lobulated, non-tender mass measuring 20 cm x 12 cm noted in the left side of her abdomen. Other systemic examinations were unremarkable. Her hematology and biochemical investigations were normal. Her urine examination showed traces of albumen. The patient underwent an ultrasound of the abdomen (USG), which revealed a well-defined complex cyst with internal echoes measuring 20 cm x 14 cm. They were discovered in the left

lumbar region, and they extended into the left hypochondrium and left iliac fossa (Figure 1). Computerized tomography (CT) of the abdomen revealed a large cystic lesion measuring 25 cm x 13 cm x 12 cm, with multiple internal radial septations with specks of calcifications noted in the left hemiabdomen. She also exhibited left mild hydronephrosis. The lesion was displacing the left kidney superiorly and laterally, and the left ureter, small bowel, and descending colon were displaced medially, suggestive of a retroperitoneal hydatid cyst (Figure 2). Her chest roentgenogram was normal, and her serological tests were negative. Echocardiography showed normal left ventricular function with an ejection fraction (LVEF) of 70%. The renal arterial Doppler showed turbulent flow in the left renal artery (peak systolic velocity (PSV): 286 cm/s; refractive index (RI): 0.46). The patient started combination therapy (albendazole (ABZ): 15 mg/kg/day; praziquantel (PZQ): 600 mg, single dose) 2 weeks prior to surgery. Three classes of antihypertensive medications were used (amlodipine: 5 mg, twice daily; atenolol: 25 mg, once daily; hydrochlorothiazide: 12.5 mg, twice daily) for adequate control of her blood pressure prior to surgery. She underwent surgery through a left flank approach, and total cystectomy was done before the abdomen was closed with a drainage tube (Figure 3). The postoperative period was uneventful. Her histopathology report revealed a hydatid cyst with no viable protoscoleces. She continued taking albendazole (15 mg/kg/day) for a 28-day cycle followed by 14-day albendazole-free interval for a total of 3 cycles. Post surgery, all antihypertensive

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Figure 1. An ultrasonogram showing a multiloculated cyst with thick internal septa in the left retroperitoneum..



Figure 2. (a) A CT showing the retroperitoneal cyst causing compression; (b) displacement of the left kidney causing mild hydronephrosis; (c) calcifications.

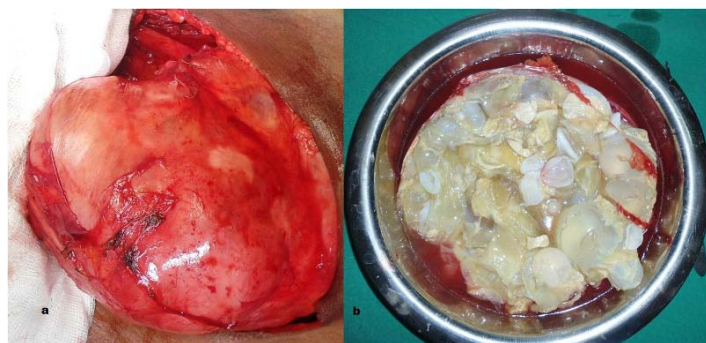


medications were discontinued and a salt-free diet was advised. She was normotensive, and there was no recurrence of hydatid disease at a 1-year follow-up.

DISCUSSION

Humans are accidental hosts for Echinococcus disease by ingesting ova through contaminated water or fomites. Once the ova are ingested, the outer protective coat is digested in the stomach. The larvae penetrate the duodenal mucosa and enter the portal circulation. Most of the larvae are trapped in the liver and the lungs; hence, they commonly noted there. Only 5 to 15% of them escape into systemic circulation [4]. Lockhart and Sapinza first reported a primary retroperitoneal

Figure 3. (a) Intraoperative imaging showing a large cystic mass; (b) the hydatid cyst after complete excision.



hydatid cyst without other organ involvement in 1958 [2]. Since then, approximately 50 cases have been reported in the literature [5,6]. In our case, primary retroperitoneal hydatid disease presented with a palpable lump, causing extrinsic compression on the left renal hilum and ureter (confirmed by radiological imaging). Secondary hypertension treated with surgery is rare and has not been reported in the literature. The cause of secondary hypertension was due to compression and the stretching of the left renal vessels and left ureter by the cystic lesion, resulting in renal hypoperfusion and obstruction, the activation of the renin-angiotensin-aldosterone system, generalized vasoconstriction, and systemic hypertension. The renal artery Doppler showed a high PSV and low renal involvement, suggestive of extrinsic compression and turbulent flow in the left renal artery. The echocardiography showed no evidence of chronic hypertension in the form of left ventricular hypertrophy. As there was extrinsic compression of the renal hilum by the cyst, further evaluation for hypertension in the form of a captopril renogram, or renal vein sampling for renin levels, was not done. Our primary aim was to control the hypertension and surgical excision of the cyst.

Three antihypertensive medications (a calcium channel blocker, a beta-blocker, and a diuretic) were given. ACE inhibitors were not given because of the increased risk of hypotension during surgery. Radiologic imaging is most important because negative serologic tests do not rule out hydatid disease. Plain abdominal X-rays may show calcification of the cyst wall. USG sensitivity ranges from 93 to 98% to detect hepatic and extra hepatic hydatid cysts. Computed tomography (CT) is the imaging technique of choice for determining the number, size, and anatomic location of the cysts, and it is also superior to an ultrasound when detecting extra hepatic cysts, as noted in our case [7]. Chemotherapy for hydatid disease is given in the form of monotherapy (ABZ) or combination therapy (ABZ and PZQ) [8]. Praziquantel is an effective protoscolicidal

agent with poor cyst-wall penetration. Albendazole is active against both protoscolecocytes and germinal epithelium. Another proposed mechanism for the enhanced efficacy of PZQ and ABZ in combination is that PZQ causes prolonged bioavailability of ABZ [9]. Combination chemotherapy was given in our case to kill viable protoscolecocytes and prevent recurrence. Surgery is curative, with cystectomy a desired option. During surgery, if complete cystectomy is not possible, partial cystectomy or marsupialization can be done after irrigating the cavity with scolicidal agents to prevent recurrence. The scolicidal agents used are 10% povidone-iodine, 0.5 % silver nitrate, 5% sodium chloride, 10% formalin, and other hypertonic solutions that prevent secondary hydatidosis. In most of the cases, complete cyst excision may be difficult because of dense adhesions or the involvement of vital organs. It is preferable to evacuate the parasite and excise the redundant portion of the pericyst, leaving the rest of the cavity open, or it can be drained externally with a wide-bore catheter. Adequate precautions should be taken to prevent anaphylaxis during surgery, which can be fatal. Recurrence is due to the rupture of a cyst, incomplete cyst removal, or previously unidentified cysts. Postoperative chemotherapy with ABZ can be given to prevent recurrence [10]. Serial imaging in follow-up care to detect recurrence is required. Although retroperitoneal hydatid disease is rare, differential diagnosis of cystic tumors like chronic abscesses, pseudo-cysts of the pancreas, chylolymphatic cysts, mesenteric cysts, soft-tissue sarcomas, or complex cysts of the kidney should be considered. Radiologists and surgeons should both be aware of this rare disease.

CONCLUSION

Ours is a rare case of primary retroperitoneal hydatid cyst presenting with an abdominal mass and hypertension. Radiological imaging was used to diagnose hydatid disease in the absence of negative serology. Combination chemotherapy was given to kill the viable protoscolecocytes and prevent recurrence if spillage occurred during surgery. Surgery cured the disease and hypertension.

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