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CASE REPORT

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Pelvic and peritoneal hydatidosis: An uncommon presentation of the common entity

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Key Clinical Message

The pelvic and peritoneal hydatidosis occurs mostly after the traumatic rupture or surgical spillage of *Echinococcus* from liver or spleen. The treatment is surgical aiming to eradicate local disease, preventing complications, and reducing recurrences.

Abstract

We report a unique case of a 26-year-old male who presented with acute urinary retention and abdominal distention. Later, CT-urography revealed peritoneal and pelvic hydatidosis behind this presentation, which was managed surgically.

KEYWORDS

echinococcosis, hydatid disease, hydatidosis

1 | INTRODUCTION

Hydatid cyst is a parasitic zoonotic disease, caused by cestode tapeworms, often due to *Echinococcus granulosus* and sometimes due to *Echinococcus multilocularis*. Humans are accidental intermediate hosts for this parasite and transmission occurs when food and water contaminated with stool is consumed from infected dogs (definitive hosts), that contains embryonated eggs. The eggs release oncospheres in intestines and these then penetrate the intestinal wall and reaches various internal organs via circulation and develop into hydatid cysts. Cysts most often involve the liver, followed by lungs, brain, bones, kidneys, and heart.[1–3] The dissemination of hydatid cyst to the peritoneum is quite unusual. Primary peritoneal hydatidosis is extremely rare (2%), and occurs via the lymphatic or systemic spread.[4,5] The seeding of Echinococcus from liver or spleen, after traumatic rupture or surgical spillage causes secondary hydatidosis.[1,6]

Here, we report a case where the patient presented with the acute urinary retention in the emergency department and was later diagnosed to have disseminated hydatid cysts in the pelvis and liver.

This case report has been reported in line with the SCARE criteria.[7]

2 | CASE PRESENTATION

A 26-year-old male presented to our emergency department with acute urinary retention for 12 h and abdominal distension for 1 day. He also complained of nonradiating pain in hypogastric region which was intermittent in nature and was associated with a single episode of nonbloody

vomiting. There was no history of any other urinary symptoms, fever, weight loss, jaundice, or breathing difficulty. He was able to pass stool and flatus. There was no history of trauma.

On examination, general condition was fair and vitals were stable. There was no icterus, edema, pallor, or dehydration. On abdominal examination, the abdomen was soft, there was a globular, nontender mobile mass of around 6 cm* 4 cm over right lumbar region extending up to suprapubic region. On digital rectal examination, the anal tone was normal, a mass of size 3 cm* 2 cm was palpable anteriorly, upper margin of which was not delineable. Other systemic examination was unremarkable.

Ultrasonography of abdomen and pelvis depicted features of bilateral hydroureteronephrosis with extremely distended bladder (around 900 mL), bilateral enlarged kidneys and grossly dilated pelvicalyceal system. A computed tomography urography was done which revealed the presence of cystic masses over the liver and in pelvic region. A well-defined hypodense cystic lesion with internal septations, of 8.3*7.9 cm size, was seen in the Morrison's pouch, anteriorly displacing and compressing the right lobe of liver. (Figure 1, green arrow) There were no signs of rupture. Three other similar lesions were also noted in the peritoneal cavity; one located posterior to the lower half of right kidney (lobulated, measuring 10 cm*6.1 cm) (Figure 2, yellow arrow); another located posterior to the bladder (measuring approximately 9.8 cm * 9.6 cm) (Figure 3, white arrow) and the third one in the left iliac fossa region (measuring 4.8 cm * 4.6 cm) (Figure 2B, green arrow). These cystic masses were displacing the adjacent bowel loops. Both the kidneys were normal in outline and attenuation, and showed prompt excretion of contrast. The patient was also found positive for Echinococcus antibody on ELISA test. Based on these findings, diagnosis of hydatid disease was made and classified as WHO-IWGE type 3a (loculated).

Urinary catheterization was done promptly, which relieved the urinary retention. Then he received albendazole 400 mg twice daily for 4 days before being operated. Exploratory laparotomy was done by giving a midline incision. Intraoperatively, we were able to see all the four cysts. We found a 7*5 cm hydatid cyst in subhepatic region; 8*8 cm and 6*5 cm cysts in peritoneal cavity, and a 10*10 cm cyst in the pelvis (Figure 4). Partial pericystectomy was done for the hepatic and pelvic cysts, wherein deroofing of the cyst with evacuation of its contents was done, while complete removal of the cyst was undertaken for the peritoneal cysts.

Subhepatic and pelvic drain were kept in place. Subhepatic drain was removed on the third day, while pelvic drain was kept for 5 days. He was then discharged on albendazole intermittent regimen for 6 months. Urine flow was normal at 2 weeks without the need of a catheter.

3 | DISCUSSION

The presentation of a patient with hydatid cyst disease is determined by the location of cysts, wherein, a disseminated disease encompasses a range of nonspecific symptoms such as abdominal pain, abdominal fullness, anorexia, and vomiting. A slow growing cyst might be asymptomatic for a variable period of time until it grows enough in size, as the clinical manifestations are due to both the number and the mass effect of the enlarging cyst. A slow growing cyst can be complicated by its rupture or rapid growth that may present as vague type of

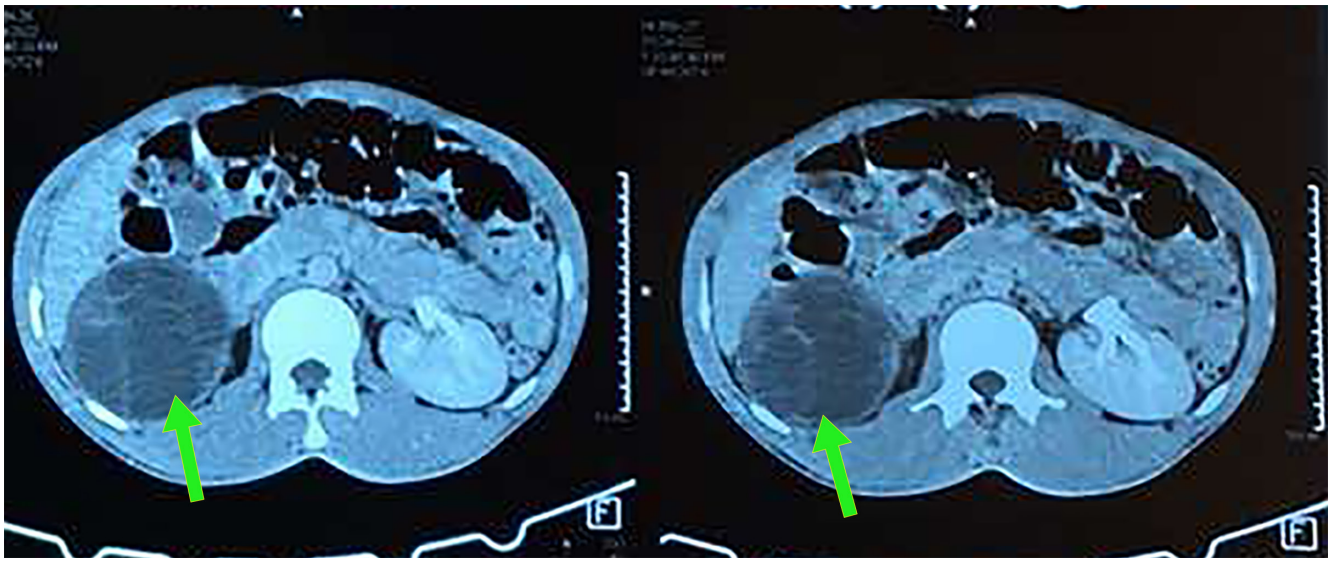

FIGURE 1 Well-defined hypodense cystic lesion with internal septations in the Morrison's pouch, green arrow.

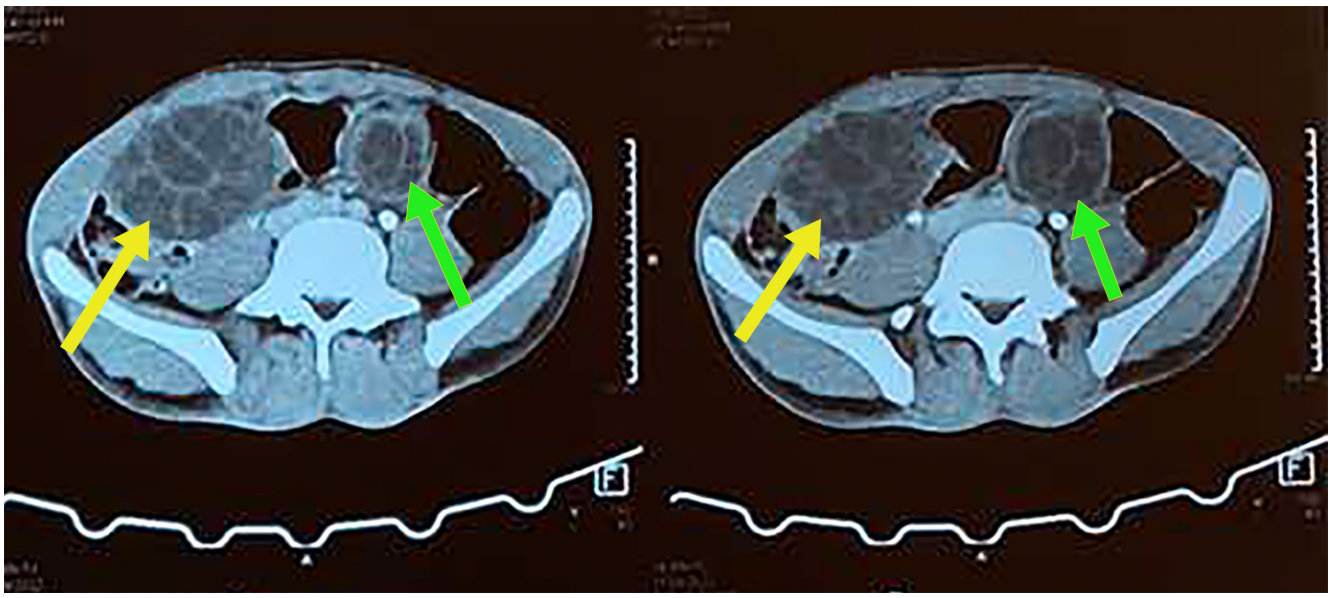

FIGURE 2 Well-defined hypodense cystic lesion located posterior to the lower half of right kidney and in the left iliac fossa region, yellow and green arrow, respectively.

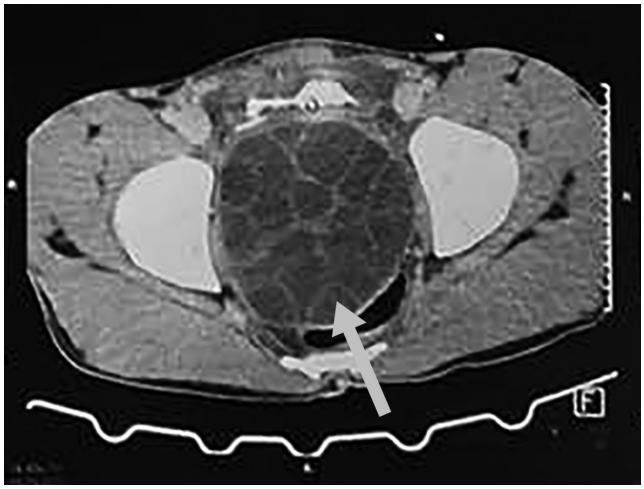

FIGURE 3 Well-defined hypodense cystic lesion located posterior to the bladder, white arrow.

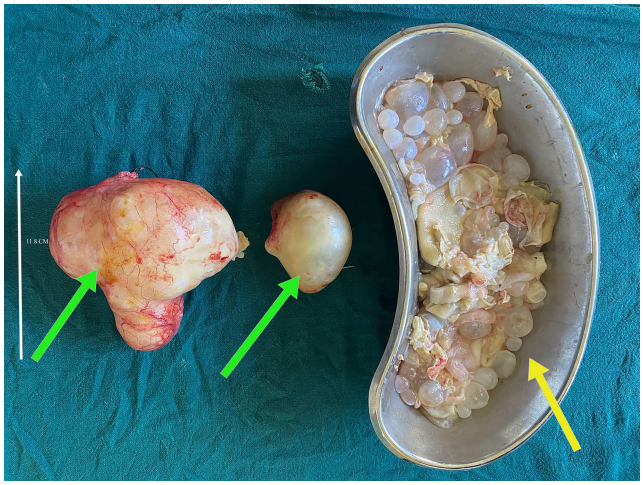

FIGURE 4 Peritoneal cysts in the left (green arrows) after total pericystectomy and multiple daughter hydatid cyst in the kidney tray (yellow arrow) after partial pericystectomy of hepatic and pelvic cyst.

acute abdominal pain.[8] Also, the released fluid may sometimes get absorbed into the circulation and cause allergic manifestations.[9]

Primary pelvic hydatid cysts are rare accounting for 2%–2.25%.[10] It manifests as a nonspecific mass with mass effect on to the adjacent organs like urinary bladder and rectum. Patients can present with acute retention of urine, obstructive uropathy, and sometimes renal failure.[11,12] Unusually, patients may present with generalized edema, a lump in the hypogastric or lumbar regions.[13]

The effective imaging modalities for the visualization of cysts for diagnosis and follow-up are abdominal ultrasound and computed tomography (CT) scan. These can be used not only for the identification of cystic stages, but also for the stage-specific approach in cystic echinococcosis (CE) clinical management. Serology testing for supporting the diagnosis can be done but there is considerable variation in sensitivity and specificity of different methods.[14,15]

Ultrasonography standardized classification of stage-specific cystic images has been issued by the WHO Informal Working Group on Echinococcosis (WHO-IWGE) for the diagnosis and the clinical management of CE. This classification is mainly based on the active-transitional-inactive status of the cyst as seen in its ultrasonographic appearance. It is useful

for decision-making and prognosis in clinical settings. Cystic echinococcosis 1 (CE1) and CE2 are active cysts containing viable protoscolices. CE3 has been subdivided into CE3a (detached endocyst) and CE3b (predominantly solid with daughter cysts). CE 4 and CE 5 are not active cysts and these cysts have normally lost their fertility and are degenerative.[16]

On the contrary, CT scan has high sensitivity and specificity in the diagnosis of the hydatid disease. Calcification and internal septa are easily detected. Laminated membrane that detaches from the pericyst is visualized as linear areas of increased attenuation within the cyst. Daughter cysts are seen as round structures found peripherally within the mother cyst.[1]

The treatment in disseminated peritoneal hydatid disease is mostly surgical. The aims of surgery are, eradication of local disease along with prevention of the complications and reduction in recurrences of the disease. Open surgery remains the treatment of choice in large symptomatic peritoneal and coexistent hepatic cysts, with good prognosis. The methods for surgical treatment of peritoneal cysts are conservative; that is deroofing the cyst, evacuation of its contents and management of the residual cavity. The ultimate goal is the total removal of the cyst, but in the case where it is not possible, partial removal of the cyst, that is, partial pericystectomy with the obliteration of the cyst cavity should be done. Similarly, in liver coexistent cysts, either a conservative approach with evacuation, partial pericystectomy as well as unroofing capitonnage pericyst suturing, cavity filling with or without drainage or radical approach with cystopericystectomy or hepatectomy is preferred.[17–19] Scolicidal agents such as povidone-iodine, praziquantel, and chlorhexidine for peritoneal irrigation, in order to prevent the recurrence of the disease. These drugs, however, are associated with their dose dependent toxicity.[20]

A concern in case of the retrovesical cyst is the reproductive structures: the seminal vesicle and the vas deferens, found adherent to its anterior wall. Adequate care must be taken to not damage these structures while removing the cyst wall. Thus, if feasible, total cystectomy is the treatment of choice. And in case it is not possible, we resort to partial cystectomy. It should be done after aspiration and sterilization of the cystic cavity with the scolicidal solution such as hypertonic saline (3%), hydrogen peroxide, and 10% formalin.[21] The administration of antiparasitic agent such as Albendazole is highly effective is the prevention of hematogenous spread of viable protoscolices.[8,22] Benzimidazoles such as albendazole should be administered after radical excision of the parasites for at least 2 years to prevent recurrences. The monitoring of the patient should be done for a minimum of 10 years.[23]

4 | CONCLUSION

Hydatid cyst is a common parasitic zoonotic disease, while primary pelvic hydatidosis is a rare manifestation of the disease that could manifest with features of obstructive uropathy as the presenting complaint. Total pericystectomy is the treatment of choice for these conditions and albendazole is used for a prolonged duration, for as long as 2 years in some cases, in the management of hydatidosis to reduce the chances of relapse.

AUTHOR CONTRIBUTIONS

Milan Kc: Conceptualization; formal analysis; investigation; resources; supervision; validation; writing – original draft; writing – review and editing. **Ishwor Regmi:** Conceptualization; investigation; resources; supervision; validation; writing – original draft; writing – review and editing. **Alok Kumar Jha:** Conceptualization; resources; supervision; validation; writing – original draft; writing – review and editing. **Biraj Pokhrel:** Conceptualization; investigation; supervision; validation; writing – original draft; writing – review and editing. **Roshan Pathak:** Conceptualization; validation; writing – original draft; writing – review and editing. **Ashutosh Kashyap:** Conceptualization; validation; writing – original draft; writing – review and editing. **Siddinath Gyawali:** Conceptualization; validation; writing – original draft; writing – review and editing. **Deepika Rijal:** Conceptualization; supervision; validation; writing – original draft; writing – review and editing. **Lalijan Awale:** Conceptualization; resources; supervision; writing – review and editing. **Abhishek Bhattarai:** Conceptualization; supervision; writing – review and editing. **Prasan Kansakar:** Conceptualization; supervision; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors declare that there is no conflict of interest regarding the publication of this paper.

DATA AVAILABILITY STATEMENT

Not applicable.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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