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CASE REPORT

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Intraperitoneal and scrotal hydatidosis: A rare case report and review of literature

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ABSTRACT

Cystic echinococcosis (CE) is a parasitic infestation caused by the tapeworm *Echinococcus granulosus*. Although adults are mostly affected, children also suffer from this disease. The lungs are the most common sites of infection in children; however, involvement of other sites is also seen. Peritoneal CE is rare in children even in endemic areas. Determining whether the origin of this site is primary or secondary remains controversial, especially in children. Secondary peritoneal CE is expected to occur after spontaneous, traumatic, or accidental rupture during surgery of primary liver or splenic cysts. Primary peritoneal CE without any evidence of another organ involvement has been rarely reported. We report a case of CE involving the peritoneum and scrotum presenting with a gradually increasing scrotal swelling in a 7-year-old child, along with sonographic, computed tomography, and MRI features.

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Abdominal pain; testicular swelling; cystic echinococcosis; ultrasound; CT scan; MRI

Introduction

Cystic echinococcosis (CE) is a worldwide zoonosis produced by the larval stage of the *Echinococcus* tapeworm. This disease continues to be a significant health problem in many endemic areas, mainly in sheep and cattle-raising areas worldwide. In endemic regions, human incidence rates for CE can reach more than 50 per 100,000 person-years, and prevalence levels as high as 5%–10% may occur in parts of Argentina, Peru, East Africa, Central Asia, and China [1]. Pulmonary location is more frequent than hepatic in children, with frequencies of 64% and 28%, respectively [2]. Other less common sites are the brain, peritoneal cavity, spleen, and heart [3,4]. Unusual sites of CE and atypical presentations in the body can be challenging. Determining whether the origin of this site is primary or secondary remains controversial, especially in children. Secondary peritoneal cysts are expected to occur after spontaneous, traumatic, or accidental rupture during surgery of primary liver or splenic cysts. Primary peritoneal CE without any evidence of another organ involvement has been rarely reported. We present a rare case of disseminated peritoneal and scrotal CE in a 7-year-old child. To the best of our knowledge, reports in the literature of isolated peritoneal CE with scrotal extension in children are very rare.

Case Report

A 7-year-old boy, resident and originating from a small village, 120 km away from Casablanca, in close contact with sheep and cattle, presented to the hospital due to an increased scrotal volume lasting several months with dull abdominal pain. There was no history of abdominal trauma, chest pain, urticaria, fever, weight loss, or anorexia nor a significant past medical history. Our patient was in a good general condition. On examination, a right hemiscrotum painless mass was identified. No signs of inflammation were found (Fig. 1). The abdominal pain was slightly exacerbated by palpation. Ultrasonography of the right scrotum showed a large well-defined cystic lesion, containing multiple rounded cysts of different sizes, echogenic foci with rounded, hyperechoic, irregular hydatid layers along with thick internal septations and daughter cysts, generating

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Figure 1. Clinical photograph of the child with scrotal swelling.

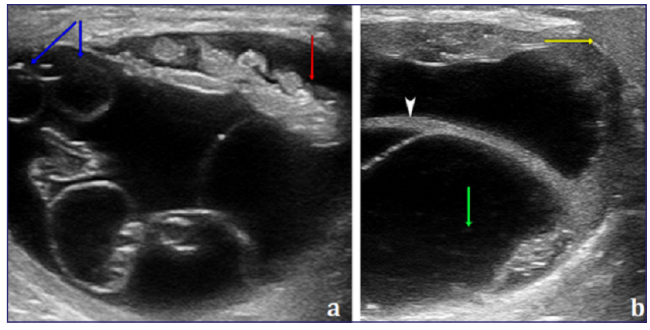


Figure 2. (a and b). Sagittal scrotal ultrasound showing a well-defined cystic lesion, containing multiple rounded cysts of different sizes, echogenic foci (green arrow) with rounded, hyperechoic, irregular hydatid layers (red arrow) along with thick internal septations (arrowhead) and daughter cysts (blue arrows), generating posterior acoustic enhancement and closely abutting the right testicle (yellow arrow).

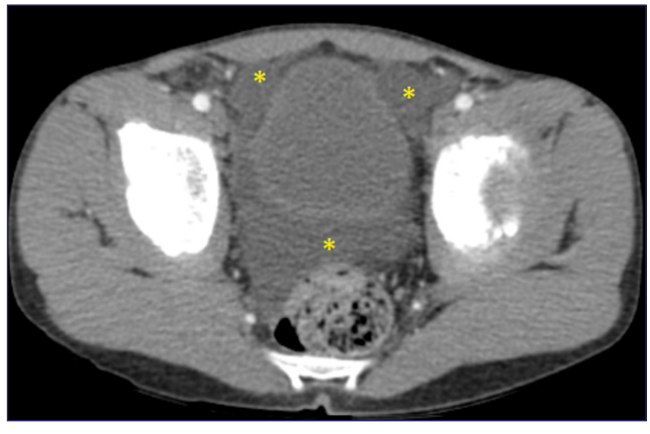


Figure 3. Axial contrast-enhanced CT scan showing multiple unenhanced intraperitoneal cystic lesions, with imperceptible walls, mainly located in the prevesical, perivesical, and retrovesical spaces (*).

posterior acoustic enhancement and closely abutting the right testicle (Fig. 2a and b). Ultrasound of the right inguinal region demonstrated a herniation of similar lesions through the inguinal canal, reaching up to the right scrotum. Contrast-enhanced computed tomography (CT) of the abdomen and pelvis revealed more in terms of the full extent of the disease. It showed multiple intraperitoneal cystic lesions, with imperceptible walls, unenhanced after intravenous administration of contrast material, filling the cavity of the peritoneum in the pelvis, mainly located in the prevesical, perivesical, and retrovesicular spaces, reaching the left and right hypochondriac regions with small amount of free fluid (Fig. 3). Right inguinoscrotal hernia through the inguinal canal, containing mesenteric fat along with cystic formations was also observed (Fig. 4). The liver and spleen were within normal limits devoid of cysts. No calcifications were observed. Further evaluation was performed with an MRI of the lower abdomen and scrotum, in order to study the relation of the cyst with contiguous organs. It showed intraperitoneal and inguinal unilocular and multivesicular cystic lesions, extended to the right scrotum, with a low-signal intensity rim on T2-weighted images, some of which containing daughter cysts as well as partially detached layers (Figs. 5–7). An additional chest X-ray did not reveal any coexistent lung localisations. Serological tests for CE were negative. Based on the typical imaging findings, the most probable diagnosis was CE.

The patient underwent a laparotomy along with a scrotal incision. Excision of the peritoneal cysts and scrotal extension without any spillage was performed (Figs. 8 and 9). After opening cyst walls, the final diagnosis of disseminated peritoneal and scrotal CE was established based on the macroscopical identification of the typical germinal layer. Postoperatively, the patient was started on albendazole tablets, 400 mg, twice a day for 3 months to prevent a recurrence. After an uneventful postoperative course, the child was discharged on the seventh postoperative day. Abdomen and scrotum were totally free from cysts with a complete absence of the free fluid during his 3 months' postoperative ultrasound check.

Discussion

In humans, the *Echinococcus granulosus cestodes* causes CE, and *Echinococcus cestodes* causes alveolar echinococcosis (AE). Although CE is common worldwide, AE is more common in Russia, Central Asia, China, Northern Japan, Central Western Europe, Eastern Europe, Turkey, and Alaska [5].

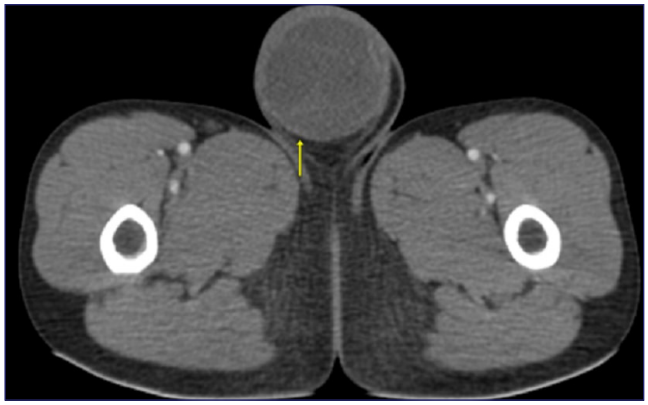


Figure 4. Axial contrast-enhanced CT scan showing the extension of the cystic formations to the right scrotum.

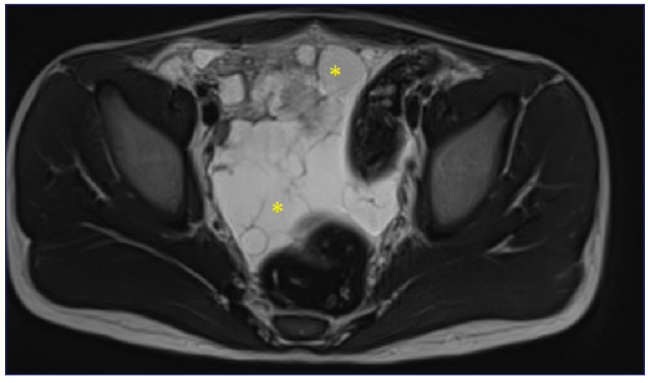


Figure 5. Axial T2-weighted MR image showing intraperitoneal multivesicular cystic lesions, some of which contain daughter cysts as well as partially detached layers (*).

CE [6] is a zoonosis produced by the larval stage of *E. granulosus*. It resides and matures in the small intestine of the dogs as a definitive host and eggs are expelled in feces. They are ingested by herbivores, often sheep, cattle, and humans. These eggs penetrate the small bowel wall and migrate to different organs [7,8]. A peritoneal echinococcosis cyst is considered primary, when no other cyst is detected [9]. It is extremely rare to have primary peritoneal echinococcosis cysts. It has an occurrence rate of 2%. Patients are usually asymptomatic for a long period of time. Echinococcosis cysts of the peritoneum are usually caused by the spontaneous or iatrogenic rupture of a hepatic, splenic, or mesenteric cyst [10]. Contamination of the scrotum would either be primary without the involvement of any other organ, or rarely secondary to rupture of an intraabdominal cyst, resulting in CE of the hernia sac as was the case in our patient. To date, only a very few cases of intraperitoneal CE with scrotal extension have been reported [11].

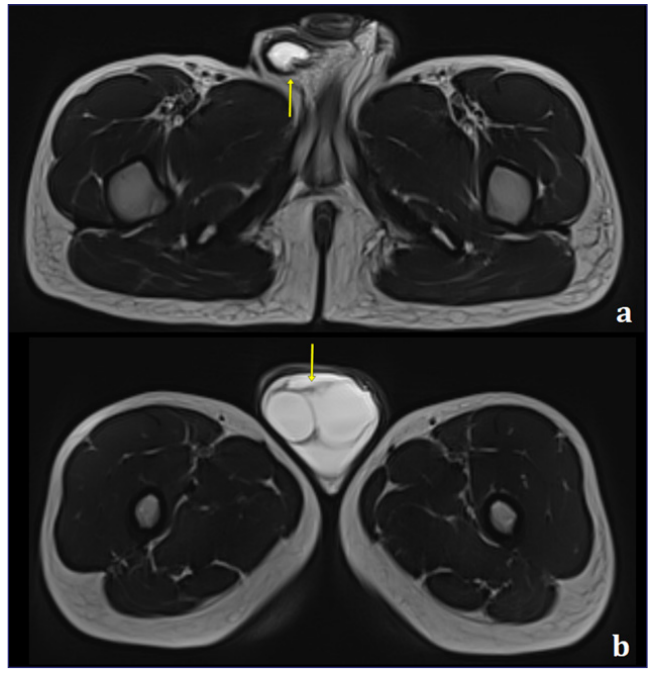


Figure 6. Axial T2-weighted MR image showing inguinal (a) and scrotal (b) extensions of cystic formations (arrows).

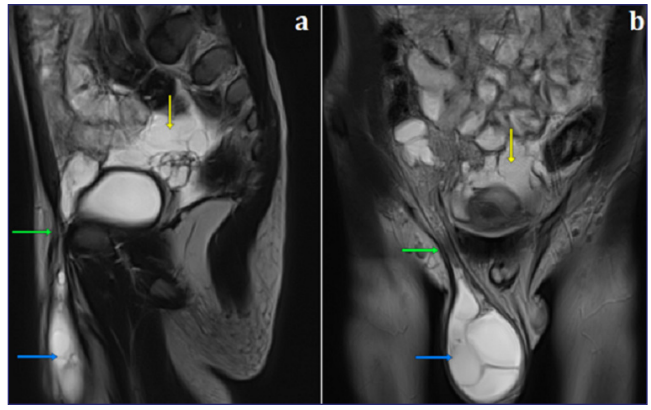


Figure 7. Sagittal (a) and coronal (b) T2-weighted MR images showing intraperitoneal cystic lesions (yellow arrows), and demonstrating herniation of similar lesions through the inguinal canal (green arrows), reaching up to the right scrotum (blue arrows).

Atypical localization of echinococcosis cysts often makes the diagnosis difficult to make. Ultrasound and CT scans have greatly improved diagnostic accuracy for the detection of echinococcosis cysts. Abdomen ultrasonography is the most common first-line radiological imaging investigation helping with determining the organ of origin and characterizing the echinococcosis cyst [12]. The typical cyst most often appears as a well-defined cystic lesion, with or without daughter cysts, with a wall formed

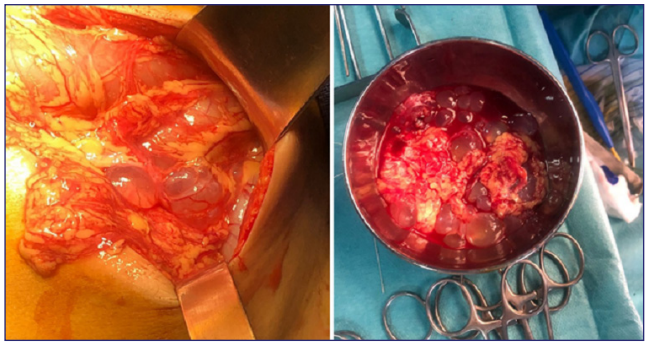


Figure 8. Intraoperative images of intraperitoneal hydatic cysts.



Figure 9. Intra operative images of scrotal hydatidosis.

by a double echogenic line separated by a hypoechogenic layer representing the germinal layer, laminated layer, and adventitial layer [6]. Simple cysts do not demonstrate internal structures, although echogenic material called hydatid sand, may be visible within the lesion when changing the patient's position [12]. Changes in the fluid pressure within the cyst, degeneration, trauma, host response, or response to therapy can lead to the detachment of the germinal layer from the laminated layer. Complete detachment of the layers leading to the collapse of the ferminal layer on a layer of fluid within the cyst manifests as the Water Lily sign. CT is indicated when US fails due to patient-related difficulties (obesity, intestinal gas or previous surgery) or disease complications. Considering that cysts may be located anywhere in the peritoneal cavity, CT remains the modality of choice, allowing imaging of the entire abdomen and pelvis and revealing more in terms of the full extent of the disease. A consecutive series of 132 patients operated on for liver echinococcosis demonstrated that CT has an overall sensitivity of 100% [13]. Contrast-enhanced CT shows well circumscribed, rounded lesions with low attenuation and no contrast enhancement [14]. Echinococcosis cyst wall typically demonstrates high attenuation at unenhanced CT. Calcification of the cyst wall could be detected on unenhanced CT. Detachment of the laminated layer from the pericyst shows hyperdense linear areas within the cyst [15]. Magnetic resonance (MR) imaging provides excellent soft tissue resolution and better delineation of the anatomy and helps with differential diagnosis. Hepatic echinococcosis cyst may have a low-signal intensity rim on T2-weighted MR images. This finding has been suggested as a characteristic sign of CE [16]. When present, daughter cysts are seen as cystic endogenous proliferation, more or less joined to the germinative layer, hypointense on T1-weighted images and hyperintense on T2-weighted images, as was seen in our case [16]. Although cyst wall calcification is more clearly depicted at CT, it has been proven that MRI seems more useful in assessing the irregularities of the rim borders, and signs leading to detachment of the layers. When it comes to collapsed layers, they tend to appear as twisted linear structures within the cyst, giving the "snake" appearance [17].

AE, on the other hand, has a tumor-like masse aspect with irregular borders, heterogeneous internal structures, and multiple distributed calcific foci. Hypoechoic/hypodense necrotic areas can sometimes be seen as large cystic cavities within the center of the lesion [5].

Imaging techniques are useful resources that allow us to navigate through the other differential diagnoses with cystic or pseudocystic masses of the peritoneum, such as peritoneal tuberculosis, mesenteric cysts, and cystic lymphangioma [18].

Several studies in turn reported that negative seroreactivity is observed in up to one-fifth of patients with CE [19,20]. Therefore, the results of serological tests should be linked to other diagnostic tests as imaging methods, and negative serological tests do not exclude a hydatid cyst infection.

The treatment aims to eliminate localized diseases, prevent complications along with minimizing the risk of recurrence. The combination of preoperative and post-operative albendazole therapy with surgery seems to be the treatment of choice to prevent recurrences, with a low recurrence rate [21]. Ultrasound and CT are regarded not only as the

main radiologic tools for establishing the diagnosis but are also invaluable in assessing the response to therapy. Being aware of the correct preoperative diagnosis is fundamental for the surgeon, it helps with being careful with an inadvertent spillage of the parasite during the operation[10].

Conclusion

In summary, peritoneal CE with scrotal extension is an uncommon entity. In countries where echinococcosis is endemic, CE should always be considered in the differential diagnosis of scrotal swelling, regardless of the presence of primary cystic lesions in the common sites. Both main diagnostic ad follow-up tools are ultrasound and CT. Surgery must be combined with pharmacological treatment to prevent recurrences.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report, including accompanying images.

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