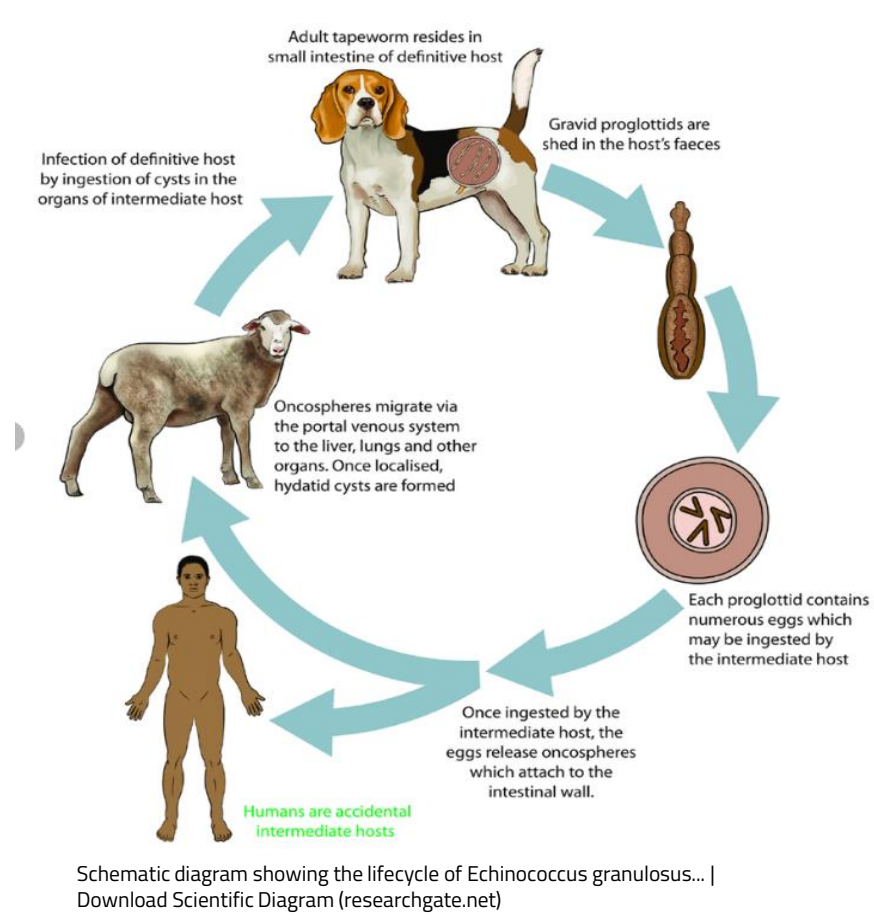


A Rare Case of Primary Pelvic Hydatid Cyst

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Introduction

Hydatid disease is a zoonotic disease caused by the parasite *Echinococcus granulosus*. While the parasite normally maintains a dog-sheep life cycle, humans can act as an intermediate host. The larvae of these parasitic tapeworms form slowly enlarging fluid-filled cysts with the potential to become very large. The disease is most prevalent in regions where humans live near host animals. Cysts have the potential to develop in any organ but are most found in liver and lungs as they typically pass through these filters early in their life cycle¹. Pelvic involvement occurs through secondary spread and rarely primary seeding. Symptoms of hydatid cysts typically depend on location and are often non-specific due to mass effect, rupture, leakage, or secondary infection. Given the rarity of primary pelvic cysts and the non-specific nature of most patient's presentations, a delay to diagnosis is common. In patients from endemic regions like Australia, although rare, primary pelvic hydatid cyst should be considered in the differential diagnosis where a diagnostic dilemma develops with a cystic lesion in the pelvis.



Case report

A 48-year-old woman presents to the emergency department with a six-month history of lower abdominal pain and two days of fevers. She described the pain as a constant, dull ache that was worse with movement. She also reported shortness of breath, dyspareunia, and abdominal bloating developing over the past 6 months with new loose stools for the past 2 days. She denied abnormal vaginal discharge, urinary symptoms, nausea, or vomiting. On history she reported a similar episode of pain 3 years ago associated with a 6cm possible ovarian cyst seen on ultrasound during an admission under general surgery for treatment of diverticulitis. She was unfortunately lost to follow up of the pelvic cyst identified at this time. She initially presented to her general practitioner with these symptoms; they referred her for an ultrasound in the community which showed a large left adnexal mass. On examination, she was febrile to 38.1°C with otherwise unremarkable vital signs. She had a soft abdomen with a tender, palpable suprapubic mass. On speculum examination, the cervix was displaced anteriorly by mass, making it difficult to visualize, though it showed no signs of inflammation, abnormal discharge or bleeding. On vaginal examination, there was a large smooth mass filling the pouch of Douglas which was tender to palpation, there was no specific uterine tenderness or cervical motion tenderness. Initial investigations revealed a haemoglobin of 136, white cell count of 15.4, normal renal and liver function, a CRP of 169, and a CA 125 of 37. A repeated ultrasound showed a 13cm large cyst at the left ovary with new concave circumferential septation with areas of avascular thickening compared to the previous external imaging. A CT abdomen/pelvis showed a large inflamed cystic mass with an internal membrane and associated bowel wall thickening. An MRI showed a large cystic mass with an internal wavy septation which appears to have delaminated from the cyst and is floating within the cyst itself. The left ovary could not be identified, and it was favored to represent an ovarian lesion. A CT chest was normal. A differential diagnosis was ovarian cyst, ovarian malignancy, tubo-ovarian abscess, diverticular abscess, primary peritoneal carcinoma, soft tissue mass, and hydatid cyst. General surgery was consulted on day one of admission due to bowel wall thickening seen on the CT. They felt the mass was unlikely to be of bowel origin with the bowel wall thickening being reactive. Infectious diseases were consulted on day 4 of admission due to persistent fevers despite broad spectrum intravenous antibiotic therapy. The patient was taken to operating theatre on day 6 of admission given persistent fevers and low risk of ovarian malignancy on all investigations to date. The intraoperative findings detailed a large pelvic mass, seen to originate from the sigmoid mesentery extending into pouch of Douglas which was adhered to the right ovary, posterior uterus and surrounding bowel. On further infectious diseases consult, it was felt that it was very unlikely to be hydatid disease given the rarity of primary pelvic hydatid cyst and that the patient showed clinical and biochemical improvement on antibiotics but echinococcus was among the serology investigations sent. The serology returned positive. On second review of the MRI images following the strongly positive hydatid serology, the imaging appearance of this cyst was indeed compatible with a primary pelvic hydatid cyst. The patient was started on albendazole and referred to general surgery for consideration of surgical management. The initial plan from general surgery was for a laparotomy + excision of hydatid cyst +/- bowel resection +/- hysterectomy +/- oophorectomy but at a follow up MRI six months after commencement of treatment, there was near complete clinical and radiological resolution. At this stage the management plan is to continue albendazole and repeat further imaging and serology in the future.

Discussion

Pelvic hydatid cysts are great imposters. The presentation is often non-specific, usually due to mass effect on surrounding organs, thus the symptoms are often misattributed. A cyst in the pelvic cavity is deemed primary when there are no other cysts identified elsewhere in the body, as was the case for this patient². More commonly pelvic cysts are secondary to rupture of primary lung or liver cysts. The incidence of primary pelvic hydatid cyst is between 0.2-2%¹. The cysts can remain asymptomatic for years until they are large enough to cause symptoms in the affected organ or they rupture causing a systemic anaphylactoid response. In Australia, the most common definitive hosts of *E. granulosus* are dogs with the intermediate host being sheep or goats. Adult *E. granulosus* reside in the intestinal tract of the definitive host. Eggs are passed to the intermediate host when the parasitic eggs are consumed, usually from pasture contaminated with dog faeces and infect the intestinal tract as well as migrate through the hematologic and lymphatic system to other organ systems. The definitive host then becomes infected by ingesting the organs of infected intermediate hosts and the life cycle continues. Humans can act as accidental intermediate hosts when they are infected by ingesting eggs, often by hand to mouth transfer from dog faeces³. The patient's history was very relevant in this case as is contained pertinent risk factors for hydatid disease. She reported previously keeping sheep and dogs on her property, the presence of wild dogs around her property, the disposal of sheep remains, and the feeding of offal to her dogs. Routine laboratory investigations are frequently normal. First line imaging for pelvic organs is typically ultrasound. Ultrasound is cost effective and can delineate features such as cyst membrane, septa, and hydatid sand within the cyst⁴. CT can help provide further information of visceral organ involvement and adherence to surrounding structures. The diagnosis is often confirmed by serological testing including fluorescent antibody and indirect haemagglutinin antibody testing. Serology is 80-100% sensitive and 88-96% specific for liver hydatid disease but this often less for other organ involvement³. The optimal treatment is pre-operative albendazole followed by surgical excision and further albendazole treatment. En bloc resection where the entire cyst is removed is the preferred surgical technique to avoid dissemination. Pre-operative treatment with albendazole can reduce cyst rupture with systemic response and re-infection⁴. Alternative treatment options include PAIR (puncture, aspiration, injection, re-aspiration) where ultrasound guided aspiration is followed injection of protoscolicidal substances and re-aspirated after a contact period of at least fifteen minutes⁴. Sole treatment with albendazole for *E. granulosus* infection has shown apparent cure in only approximately 30% of cases⁵. These alternative options are considered for patients with multiple large cysts in the liver, spleen, bones, inoperable cases, or recurrence following surgery⁴. Liver function abnormalities are common but rarely serious, while infrequently haematological changes affecting white cells can be more serious, thus patients should be monitored during treatment⁵. In this case, the patient had complete resolution with albendazole only. Preventive measures include regular prevention for dogs to break the life cycle, especially dogs kept around sheep farming, avoiding feeding offal to dogs, hand hygiene, and washing of vegetables.

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Hydatid Serology

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