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Primary Hydatid Cyst Diagnosed as Ovarian Neoplasm- A Case Report

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Abstract

Introduction: Primary hydatid cyst is rarely located in the pelvis. It can be easily misdiagnosed as ovarian neoplasm leading to inadequate preparation for the surgery. Main symptoms and important findings: Seventy nine years old postmenopausal lady presented with abdominal pain and difficulty in urination for four days. Her CA-125 levels were raised and abdominal imaging revealed large complex multiloculated ovarian mass causing bilateral gross hydroureteronephrosis along with left ureteric calculus.

Therapeutic Intervention: She underwent exploratory laparotomy, removal of the mass and open uretero-lithotomy under general anesthesia, but sustained cardiac arrest in the post-operative period and succumbed to it. The mass was sent for frozen section during surgery, which revealed the diagnosis of hydatid cyst.

Conclusion: The risk of morbidity and mortality in a case of pelvic hydatid cyst can be minimized by pre-operative suspicion along with careful dissection and appropriate postoperative management.

Keywords: Hydatid cyst in pelvis; Differential diagnosis of ovarian neoplasm; Ovarian mass in old women; Primary hydatid cyst

Introduction

Hydatid disease, or Echinococcosis, is a parasitic infection caused by larval stage of *Echinococcus granulosus*. It is endemic in cattle grazing regions of the world like Mediterranean, Middle East, Eastern Europe and South America [1]. It usually involves the liver and lungs. Although it may attack all the organs, its location in the pelvis is very rare. It is usually due to the rupture of cyst in other areas of the body [2]. Only 12 out of 532 cases of pelvic hydatid cyst were found over a period of 20 years in an endemic area [3]. In this case report, we present the rare occurrence of a primary pelvic hydatid cyst in an octogenarian woman, which manifested itself with urinary symptoms and was misdiagnosed as an ovarian neoplasm and the patient succumbed in the post-operative period. This case emphasizes the importance of suspecting pelvic hydatid cyst in a case of complex pelvic mass, along with the significance of careful intraoperative dissection as well as appropriate postoperative management.

Patient Information

Seventy-nine years old postmenopausal lady presented to the hospital with complaints of abdominal pain and difficulty in passing urine for last four days. There was no history of any bowel disturbance. She did not give history of loss of weight or appetite. There was no history of fever. She was postmenopausal for more than 35 years and was a known hypertensive for four years. She had a vaginal delivery 50 years ago. There was no history of any surgery in the past or tuberculosis or malignancy in her family. She was a homemaker, living with her children and hailed from a rural area. She belonged to a lower-middle socioeconomic status and had never attended school.

On general physical examination, the patient was moderately built and nourished. She was afebrile, Blood Pressure (BP) was 140/80 mmHg and, pulse rate was 82 beats/min. The patient's respiratory, cardiovascular, and neurological systems were normal. The abdominal examination revealed 2 cm × 2 cm paraumbilical hernia. Abdomen was soft on palpation, with mild diffuse tenderness, but no rigidity. There was no hepatosplenomegaly or ascites. A suprapubic hard, fixed non tender mass was felt arising from the pelvis about 4 cm above the pubic symphysis corresponding to the size of 14 to 16 weeks of the gravid uterus. There was no lymphadenopathy. On auscultation,

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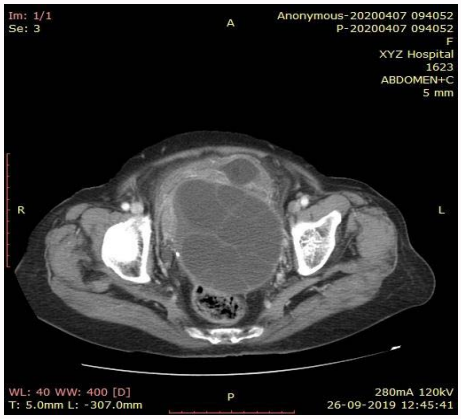

Figure 1: Ovarian mass as seen on CECT.

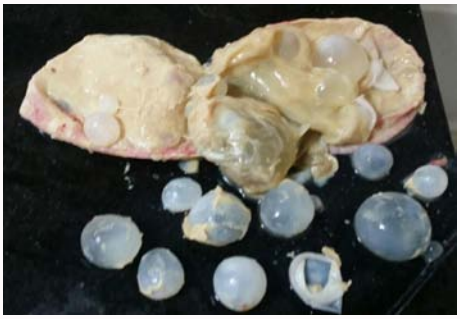

Figure 2: Ovarian mass showing daughter cysts inside.

bowel sounds were normally heard. Bimanual examination revealed a 14 cm × 12 cm non tender, hard, immobile mass with a smooth surface. Mass was impacted in the pouch of Douglas and felt through all vaginal fornix, not separate from the uterus. The rectal examination confirmed the findings.

The patient was catheterized, and Ultrasonography (USG) of abdomen and pelvis revealed 12 cm × 12 cm mass arising from left ovary suggestive of serous cystadenoma or cystadenocarcinoma. There was gross hydroureteronephrosis bilaterally. She was found to have high blood sugars and deranged renal functions. Contrast Enhanced Computed Tomogram (CECT) of abdomen and pelvis, as seen in Figure 1, showed multiloculated complex cystic lesion, likely to be arising from left ovary, posterior to urinary bladder displacing the urinary bladder and the rectum and causing bilateral hydroureteronephrosis. The uterus was displaced laterally to the right side, along the right lateral margin of the lesion. Superiorly, the lesion reached up to the lower border of L5, and the lesion was abutting the L5S1 disc. Right renal calculus and left distal ureteric calculus of size 1.5 cm was noted along with left paraumbilical hernia. Other abdominal organs were normal, and there was no free fluid in the abdomen. The serum CA-125 level was 179 U/ml. Cystoscopic removal of ureteric calculus was attempted but failed due to as both the ureteric orifices could not be visualized. She underwent exploratory laparotomy through a midline vertical incision, and open uretero-lithotomy under general anesthesia after optimizing BP and blood sugars.

Intraoperatively, well-defined cystic lesion of size 12 cm × 12 cm × 8 cm was noted impacted in the pouch of Douglas, densely adherent to the posterior wall of uterus and displacing the uterus to the right with the left fallopian tube stretched over the mass. Both ovaries were normal. The lesion was densely adherent to the anterior surface of the rectum and sigmoid colon. The capsule was intact, and there was no free fluid in the pelvis. The pelvic lymph nodes appeared normal in size and echotexture. No other intraabdominal pathology was found. The liver was explored and no lesions were noted. There was no lesion in the omentum, small and large bowel. The mass was very slowly dissected out in to from the surrounding structures with sharp dissection with great difficulty and required a lot of patience and time. After removal, it was sent for frozen section where it was cut open as seen in Figure 1. Frozen section revealed the diagnosis of hydatid cyst with congested blood vessels on the attached peritoneum its surface and multiloculated grape- like cysts as seen in Figure 2, with pultaceous material in the inner cyst wall, and plenty of scolices with hooklets of *Echinococcus granulosus* in the laminated cyst wall. As the diagnosis of ovarian malignancy was excluded, omentectomy and pelvic and paraaortic lymphadenectomy were not done. After removing the lesion, left uretero-lithotomy and ureteral Double J (DJ) stenting was done to remove the left ureteric calculus. Peritoneal lavage with 0.9% Sodium Chloride (NaCl) and irrigation of the cavity was done with 3% NaCl solution. She received two units of packed cells intraoperatively to combat hemorrhage. Due to the chronic nature of the mass leading to dense adhesions with its surrounding structures, the surgery lasted for 8 h for its removal in to. Three hours after the surgery, the patient sustained cardiac arrest and was revived with one cycle of Cardio-Pulmonary-Resuscitation (CPR). Gradually, she developed respiratory distress, hypotension, metabolic acidosis, and acute kidney injury and succumbed to it, despite the maximal supportive care. The final histopathological diagnosis was hydatid cyst involving the pouch of Douglas.

Retrospectively obtained, the patient did not have any history of travel to the endemic area or any association with dogs or pets anytime. The relatives denied the history of the patient being in the farm in the past.

Discussion

Echinococcus granulosus is a parasite in which dog acts as the definitive host where it completes its life cycle, and cattle, sheep, goats, or pigs act as intermediate hosts. Human is an accidental host. Humans get infected after ingesting eggs, excreted from the feces of dogs. The eggs then hatch in the intestinal tract to form oncospheres, which pierce the intestinal wall and spread via blood and lymphatic circulation to various organs, where they grow to form hydatid cysts. Hydatid disease is usually acquired in childhood, though the symptoms appear 5 to 20 years after exposure when the diagnosis is made. The dense adhesions around the cyst, in our case, suggest its chronicity. The patient most likely got infected by ingesting the ova with unwashed vegetables or association with pets when she was young. Also, the ova remain viable for a long time, causing the delayed transmission to humans who have no direct contact with vector animals.

Hydatid cysts are prevalent worldwide, the highest being in the Middle East, Australia, Argentina, New Zealand, Africa, and the Mediterranean region [1]. The worldwide prevalence has decreased considerably over the last few decades [4]. Some parts of north India also show high prevalence of hydatid cysts. The annual incidence varies from ≤ 1 to 200/100,000 populations [5]. Epidemiological data from India are scarce. A study from the eastern part of India found female preponderance with the median age of presentation at 33 years [6].

The female preponderance may be mainly due to their involvement in farming and animal breeding. Ignorant of the importance of hygiene, as well as of the fact that, eating contaminated food can spread the disease, is mainly responsible for the transmission of infection in India [7]. Factors such as poor socio-economic conditions and lack of education, add to the disease's burden.

Liver is the most common site for Echinococcal cysts (60% to 70%), lungs (10% to 25%), followed by the spleen, kidney and, brain [8]. Primary peritoneal hydatid cysts are very rare [9]. Pelvic location is very uncommon, as seen by Gazzar et al. [10], who found only one case of hydatid cyst in the pelvis, out of 51 cases in a span of 4 years [10]. Pelvic hydatid cysts can cause abdominal pain, lump, menstrual irregularities and infertility in younger women, and pressure symptoms [11]. Pelvic Echinococcosis may resemble malignancy as happened in our case, which mimics multicystic ovarian neoplasm [12].

Our patient presented with a large pelvic mass with chronic lower abdominal pain and pressure symptoms on the urinary tract causing bilateral hydroureteronephrosis. Her advanced age, and imaging showing large complex multiloculated cystic mass with septations, along with raised CA-125 levels, raised the possibility of an ovarian neoplasm. The diagnosis of hydatid disease was never suspected because of its rarity, absence of typical epidemiology (endemic area, association with dogs and cattle), and absence of similar lesion at other sites in the abdomen.

Correct preoperative diagnosis of Echinococcosis is very important, as specific precautions are required to prevent dissemination and seeding of the surgical field. However, as the diagnosis was never suspected in our patient, the recommendation of an image-based stage-specific approach for Cystic Echinococcosis (CE) by Brunetti et al. [13], for the World Health Organization Informal Working Group on Echinococcosis (WHO-IWGE) was not followed [13]. Anaphylactic shocks resulting from intraoperative spillage after a mistaken diagnosis of a retroperitoneal tumor leading to death have been reported. According to WHO-IWGE report, there is no 'best' treatment for CE due to the absence of evidence involving clinical trials with different modalities [13]. Total cystectomy is the treatment of choice for symptomatic hydatid cysts [14]. However, consensus by the WHO-IWGE experts is that adequate therapy should be based on image-based staging. An alternative therapeutic approach for densely adherent peritoneal cysts can be uncovering and draining of the cyst. The abdominal cavity is isolated with gauzes soaked in 20% saline to prevent the spread causing hydatidosis, and allergic reaction [15]. The most common complications are accidental rupture and secondary infection, which can be reduced by albendazole given preoperatively [1,12]. Another alternative therapy involves using concomitant anti-helminthic drugs. In this therapy, percutaneous aspiration of the cyst is done under USG guidance, followed by the injection of protoscolicidal substances (20% NaCl solution, 95% ethanol, or betadine solution). The cyst is totally removed after 30 min of instillation. In our case, although there were dense adhesions requiring time and patience, the cyst was dissected surgically without rupture.

The incidence of hydatid cysts in the pelvis is very low [9]. However, cases are being reported from non-endemic areas, which present a challenge in the diagnosis, as in our case. Hence, primary pelvic hydatid cyst should be suspected while evaluating a case of cystic mass in the pelvis. Preoperative suspicion, careful intraoperative dissection, and postoperative care can help in appropriate management without causing much morbidity or mortality which was seen in our case.

Ethics Approval

Ethical approval was obtained from the Institutional Ethics Committee for publication of the case.

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