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Case report

Primary hydatid disease of fallopian tubes: the risk of infertility looms large

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ABSTRACT. A 45-year-old postmenopausal woman from a rural area presented with a painless lower abdominal lump. Contrast-enhanced computed tomogram of abdomen revealed a well defined hypodense pelvic cystic lesion with multiple daughter cysts suggestive of hydatid disease. The liver was free of cysts on imaging. On laparotomy, the cyst cavity was found to be communicating with the fimbriated ends of both the fallopian tubes. Cyst excision along with hysterectomy and bilateral salpingo-oophorectomy was performed. Histopathology confirmed presence of hydatid disease by demonstrating daughter cysts and laminated membrane completely filling up the tube lumens. The uterus and ovary were uninvolved. While the management is straightforward in postmenopausal women, the risk of infertility looms large in young patients with bilateral fallopian tube hydatid disease. Proper preoperative counseling is thus essential in the patients with pelvic hydatid disease to safeguard against future litigations.

Keywords: fallopian tube, hydatid, pelvic, fertility, albendazole

Introduction

Hydatid disease (HD) or cystic echinococcosis is a zoonotic disease caused by the parasite *Echinococcus granulosus*. The life cycle of the parasite extends between a primary canine host such as dog and an herbivore intermediate host like sheep or cattle. Humans act as accidental intermediate hosts when the parasite gains entry through ingestion of food contaminated with echinococcal eggs. The *in vivo* spread of the parasite is primarily hematogenous with liver and lungs being the most commonly affected organs. Involvement of other organs such as spleen, kidney, brain, heart, bone, peritoneum, and reproductive system is encountered in only 5% of patients [1]. Amongst the reproductive organs, involvement of the ovaries and uterus are more frequent than the cervix and fallopian tubes. Primary involvement of fallopian tubes is an extremely rare event [2]. We herein, report our experience in the management of such a patient with primary HD affecting both the fallopian tubes.

Case presentation

A 45-year-old multiparous woman from a rural area presented with a painless progressive lower abdominal lump of 12 months duration. The patient had neither bowel nor urinary complaints. Her appetite was normal with no recent loss of weight. She had entered menopause two years back. A solitary nontender, firm and immobile lump of 15×8 cm size was felt occupying the hypogastrium and umbilicus region on abdominal palpation. Smooth anterior rectal wall bulge was appreciated on digital rectal examination. Per vaginal examination was unremarkable. Ultrasonogram showed a hypo-echoic cystic lesion with fine internal septations suggestive of pelvic HD. Contrast enhanced computed tomogram (CECT) of abdomen demonstrated a well-defined hypodense lesion of 19.4×18.4×12.3 cm size occupying the pelvis and lower abdomen. Presence of multiple rounded cystic structures of variable sizes (daughter cysts) clinches the diagnosis in favor of HD on CECT. The uterus and adnexal structures were not separately identified from the large cystic lesion. The liver was

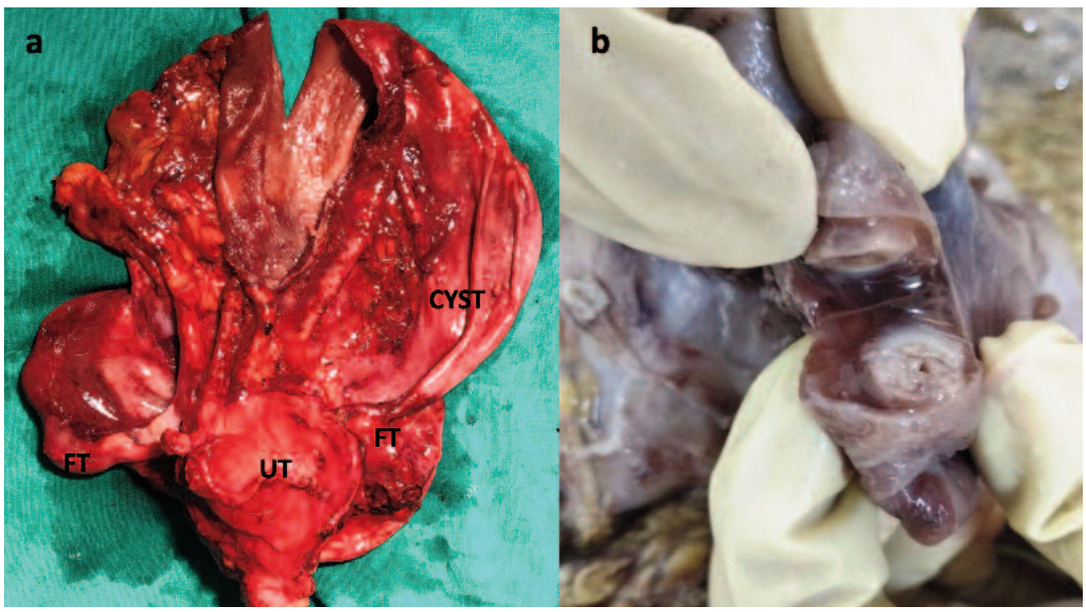

Figure 1. a: resected specimen showing the densely adherent hydatid cyst to the uterus (UT) and both fallopian tubes (FT); b: lumen of the fallopian tube filled with laminated membrane of the hydatid cyst

devoid of cysts in the CECT scan. Serology of the patient was positive for echinococcal antigen. She was subjected to laparotomy following 30 days course of albendazole monotherapy. On exploration, the cyst was found to be densely adherent to the posterior wall of uterus, both ovaries, and fallopian tubes. The cyst contents were aspirated followed by instillation of hypertonic saline into the cyst cavity. Communication between the cyst and both fallopian tubes was identified on deroofing the cyst cavity. Excision of the cyst along with total hysterectomy and bilateral salpingo-ophorectomy was performed. On histopathology, lumen of both the fallopian tube were packed with daughter cysts and multiple fragmented lamellar membrane of HD (Fig. 1). The uterus and the ovaries were found to be uninvolved in the disease process. Postoperative recovery was uneventful and the lady was free of recurrence at 6 months follow up.

Discussion

Hydatid disease is prevalent in countries having a sizeable rural community dependent on agricultural income. Close proximity to dogs and livestock is known to increase the risk of contracting the disease. The indolent nature of the parasite accounts for the major chunk of patients reporting late in an advanced stage of the disease. Hydatid disease involving the female pelvic cavity often masquerades the clinic-radiological picture of complicated ovarian cyst, dermoid cyst, cystic ovarian tumor, and pelvic abscess [3]. In view of nondescript clinical manifestation, the possibility of pelvic HD should be considered in patients hailing from endemic areas, history of prior treatment for HD, positive serology for echinococcal antigen, range bound tumor marker levels, and characteristic CECT findings such as presence of daughter cysts along with floating laminated membranes and multiple septa [4]. Preoperative diagnosis of HD is extremely useful as it mandates initiation of prophylactic albendazole therapy and alerts the surgeon against intra operative spillage of contents to curtail the risk of recurrence and anaphylaxis.

Majority of pelvic HD are secondary to spontaneous or surgical rupture of liver HD. Primary pelvic HD is rare in women with a reported incidence of 0.2–0.9% [5]. Hematogenous or lymphatic dissemination and direct transmigration of hexacanth embryos through the bowel wall are the proposed mechanisms for the occurrence of pelvic HD sans liver HD [6]. The clinical presentation is also different in both types of pelvic HD. The patients with secondary pelvic HD are more likely to suffer from anaphylactic reactions, superadded infections and rapid disease progression due to sudden release of a large parasitic load following liver HD rupture. In contrast, slow contamination by relatively fewer parasites in primary pelvic HD is characterized by delayed and subdued clinical presentation. The deep recess of

cul-de-sac acts as the stomping ground for the parasitic embryos gaining an entry into the peritoneal cavity. The adjacent uterus and adnexal structures are subsequently involved though extension of this cul-de-sac infection. Identification of laminated membrane and daughter cysts inside the fallopian tube lumen without uterine involvement in our patient points to retrograde extension of cul-de-sac infection through the open fimbriated ends of the tubes.

Surgery remains the treatment of choice in patients with symptomatic pelvic HD. The goal of surgery is complete removal of local disease, preservation of organ function, and elimination of future risk of recurrence. Surgery in reproductive organ HD should be tailored according to the age of the patients. Conservative cyst excision with preservation of fertility should be the focus in young women while total hysterectomy with bilateral salpingo-ophorectomy should be reserved for postmenopausal women. Salpingectomy is considered appropriate surgical treatment for unilateral tube involvement. Management is tricky in bilateral disease as excision of both the tubes will jeopardize the prospect of future fertility in patients who are yet to complete their family while pharmacological management alone will be ineffective in clearing the blockage of the tubal lumen by the daughter cysts and laminated membranes. Hence the risk of infertility should be discussed beforehand with the young women heving pelvic HD to avoid future litigations.

In conclusion, pelvic HD with bilateral fallopian tube involvement is a dreadful condition adversely affecting the fertility of young patients. Modern imaging modalities often fail to detect fallopian tube involvement in patients having pelvic HD. A high index of clinical suspicion is thus essential for early diagnosis and institution of prompt treatment in these patients. At the same time proper counselling regarding future risk of infertility is absolutely essential in all the patients with pelvic HD.

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