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Rare Multiple Recurrences of Multivisceral Echinococcosis

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1. Abstract

Cystic echinococcosis (CE) is a kind of globally distributed hydatid disease, which is caused by the parasite Echinococcus, and it is still an important public health concern in less developed countries. This article reported a rare case of pelvic and retroperitoneal echinococcosis, with a history of multiple recurrences of liver and lung echinococcosis.

2. Introduction

Cystic hydatid is a zoonotic disease caused by the larval stage of echinococcus. People who live with sheep, cattle, goats, camels and horses, are more likely to develop cysts after they accidentally eat the parasite's eggs as an intermediate host. The liver and lungs are the most commonly affected organs, but virtually any organ can be affected. The multivesical hydatid cyst case involving retroperitoneum and pelvis is rarely reported, is usually secondary to the rupture of liver cyst, but can also be manifested as primary lesions affecting the reproductive organs [1]. Multivisceral echinococcosis is defined that hydatid cysts occur in more than one organ at the same time, which is rarer than multiple echinococcosis that means multiple hydatid cysts occur in the same organ [2]. The treatment is anti-parasitic chemotherapy (Albendazole) combined with surgery. It is a challenge in the clinic to reduce the recurrence rate of this disease.

3. Case Presentation

A 46-year-old Chinese woman living in a rural area of Gansu province, complained of pelvic masses discovered five months later after hepatectomy for hydatid disease. Anorexia, cough, fever, abnormal vaginal bleeding, abdominal pain and weight loss did not happen. The woman had close contact with dogs and sheep during her childhood, was diagnosed with hepatopulmonary echinococcosis, and underwent her first liver and lung echinococcosis resection 27 years ago without anti-hydatid drug treatment. Ten years later, she suffered abdominal pain again and was diagnosed with recurrent hepatic echinococcosis. Then, she underwent a second liver resection. One year ago, the same thing happened again and she underwent her third liver resection. Besides, she was diagnosed with hepatitis C and treated. However, there was no exact etiological factor found for the hepatitis C. At present, B-ultrasound showed two large honeycomb-shaped cystic masses in her pelvis, and one was in the right anterior part of the uterus, with about 82*72mm in size (Figure 1A), the other one was in the left posterior side of the uterus, with about 79*66mm in size (Figure 1B). Considering that the patients was in the recovery stage after the third surgery, it was firstly recommended to take 200mg albendazole twice a day, and receive surgical treatment afterwards. There was a 30cm curved scar from the right upper abdomen to the right back, a 10cm longitudinal scar in the middle of the abdomen, and a 13cm transverse scar on the lower abdomen. Bimanual examination revealed a 6*5cm cystic mass in the right adnexal region, occupying the Douglas pouch and pulling the uterus to the right back with limited mobility. No other abnormalities were found obviously in the systemic and gynecologic examination.

After admission, pelvic ultrasonography detected two irregular mixed-echo masses in the right adnexal area, the size of which were 56*57mm and 38*24mm, respectively, showing that there was no vascularity in the mixed-echo masses with solid components (Figure 1C). CT scan showed that the Douglas pouch was closed and some irregular nodules were observed in the right rectal fossa, and the rectum was compressed and deformed (Figure 2A and B white arrows). A cystic solid mass, with the size of 54*53mm, was visible above the front of the nodule and the density of the solid part was uneven (Figure 2A, B red arrows). It was demonstrated that the lesion was closely related to the right inter-

nal iliac arteries and veins by Computed tomography angiography (CTA) (Figure 3A and B).

After multidisciplinary consultations with gynecology, hepatobiliary, anesthesiology, imaging and gastrointestiny, a comprehensive assessment of the patient's condition was conducted, and a laparotomy was arranged. Before the operation, according to the Chinese local echinococcosis treatment guidelines [3], the dosage of Albendazole was adjusted to 400mg twice a day (10~15mg/Kg/day) for 1 week. 100mg hydrocortisone was given before surgery to prevent possible allergy. The operation revealed extensive adhesions in the abdomen. There were no obvious abnormalities in bilateral ovaries and fallopian tubes. There were multiple gray-white, thick-walled cystic masses in the pelvic cavity, and the largest one, approximately 6cm, was located in the right vascular area, close to the back of the broad ligament, and its basement was adhered to the right ureter (Figure 4A and B). There was a 2cm mass on the left side of the posterior wall of the uterus near the cervix (Figure 4C), and the other three lesions, about 20~40mm, were located in the Douglas pouch and the retroperitoneal space. The hydatid cyst was carefully aspirated and the cyst wall was excised along with the clearness of the involved area. Adjacent soft tissues were extensively removed, and hysterectomy and bilateral salpinggo-oophorectomy were also performed. The area was washed and soaked with 10% NaCl solution several times before and after section to avoid leakage of the cystic fluid. The gross morphology of the cystic lesion resembled a multilobulated ovoid mass (Figure 5A) and laminated membrane and scolices surrounded by fibrous capsule and atrophic myometrium under the microcopy HE×40 (Figure 5B). The patient continued to take Albendazole 400mg twice a day(10~15mg/Kg/day) after operation to prevent recurrence. One year after discharge from the hospital, pelvic examinations, CT and B-ultrasound showed no sign of recurrence.

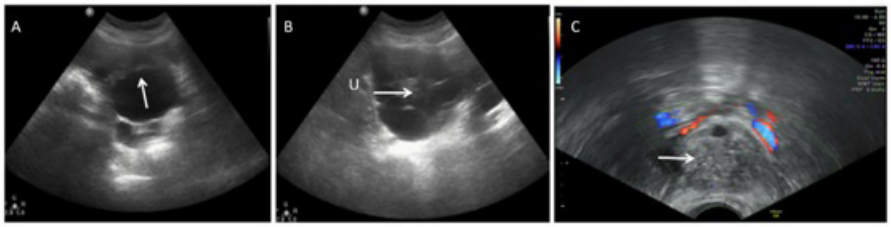

Figure 1: Ultrasound image showing two large honeycomb-shaped pelvic cystic masses, **A** one is in the right anterior part of the uterus, about 82*72mm in size, **B** the other one is in the left posterior side of the uterus (*u*), about 79*66mm in size, **C** Transvaginal Color Doppler of the pelvis showing the mixed-echo mass without vascularity in the solid component (*White arrows*).

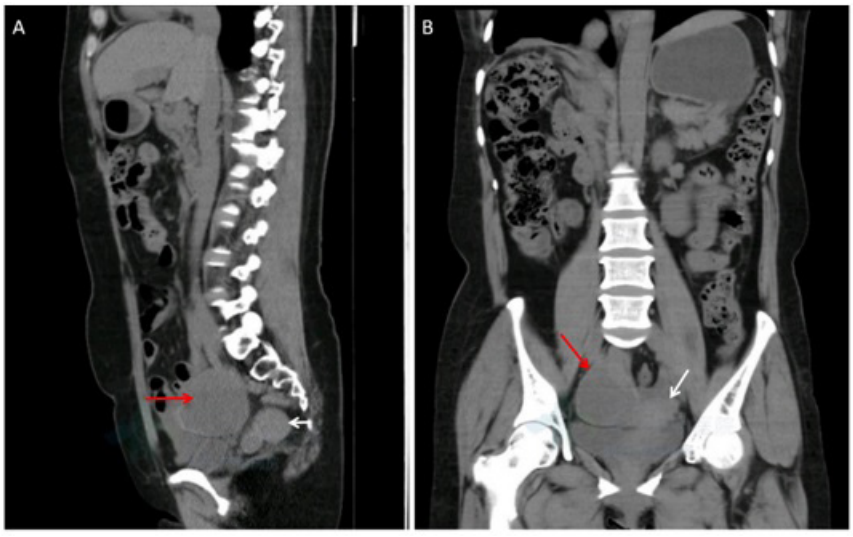

Figure 2: A Coronal and **B** Sagittal CT images showing the closed Douglas pouch and some irregular nodules on the right rectal fossa, compressing the rectum (*white arrows*). A cystic solid mass above the front of the nodule, 54*53mm in size, with uneven density of solid part (*red arrows*).

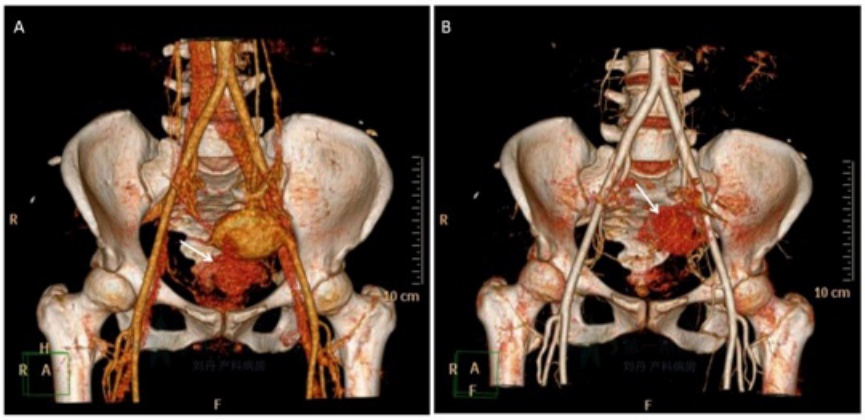

Figure 3: A Arteriography and **B** Venography showed that the lesion was closely related to the branches of the right internal iliac artery and vein (*White arrows*).

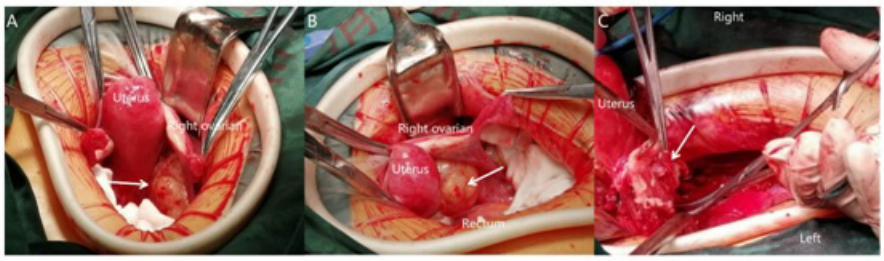

Figure 4: A, BOperative vision of pelvic multivisceral Echinococcosis, the largest echinococcus cyst was located in the right vascular area (Front and side view), C elimination of the daughter cysts of lesion near the cervix on the left side of the posterior wall of the uterus (White arrows marked the hydatic cyst).

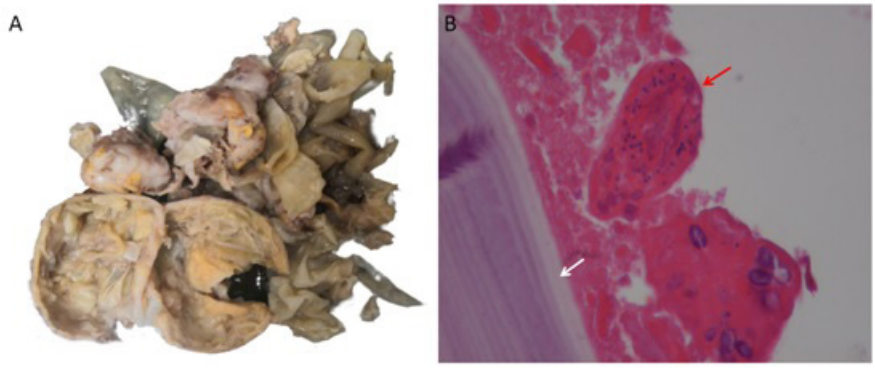

Figure 5: A the general appearance of the hydatid cyst showed the cyst capsule contained numerous daughter cysts, **B** laminated membrane (*white arrow*) and scolices (*red arrow*) of the hydatid cyst (*HE, original magnification x40*).

3. Disscussion

Human hydatid disease or echinococcosis, is an uncommon but serious parasitic zoonosis, mainly caused by the tapeworm *E. granulosus*, and it is especially popular in countries where sheep and cattle are intensively herded, such as South America, the Middle East, Sub-Saharan Africa, Central Asia and northern Canada. Due to frequent overseas travel, it may appear anywhere in the world today [4].

Echinococcus cysts can occur in any part of the human body, with the liver (75%) and lung (24%) mostly [5]. The occurrence of pelvic echinococcus cysts is 0.2-0.9% [6]. About 80% of primary pelvic lesions involve the reproductive organs, of which the most common site is the ovary, followed by the uterus that is even rarer [7]. Theoretically, it could affect any part of the pelvis, including the intestines, rectum, bladder, broad ligament, pouch of Douglas, retroperitoneum, and even bones and joints, which are usually secondary to accidental or spontaneous rupture of cysts in other areas, accompanied with adhesions of the omentum to the cyst wall and multiple adhesions between the cyst, the bowel, bladder and other pelvic organs [7]. The woman had multiple organs involement, and hepatic echinococcosis, pulmonary echinococcosis, pelvic and retroperitoneal echinococcosis and extensive abdominal adhesions were observed during the surgery. Cyst formed in the pelvis may be asymptomatic for a long time and be discovered accidentally when complications (rupture of the cyst, infection, mechanical compression of adjacent organs, etc.) cause symptoms [8]. Whether the disease has developed to the middle or late stage depends on the cyst size, location, space occupied by the cyst and the degree of compression on adjacent tissues or organs [9]. Hydatid disease is extremely rare in the female reproductive tract. When it comes to the female reproductive system, some unusual and interesting

obstetric and gynecological symptoms might occur, such as vague lower abdominal pain or swelling, irregular menstrual cycle, secondary infertility and stress symptoms like renal and ureteral hydrops. These factors should be considered in the differential diagnosis of uterine myoma or ovarian malignancies. For pregnant women with echinococcosis, childbirth may be complicated by hydatid cysts either with failure of pregnancy failure or obstruction of delivery [7]. For diagnosis, clinical history and symptoms, serodiagnostic techniques such as serological detection of specific immunoglobulin G (IgG) antibodies, radiological examination, and histopathologic analysis of the biopsy specimens are very important [10]. The microscopic examination which could show the hooklets and scolices within the laminated cyst is regard as the gold standard. Ultrasonography is the preferred method to check and evaluate the number of hydatid cysts. Surgical resection is the optimal treatment combined with preoperative albendazole treatment at a dose of 10-15 mg/Kg/day to sterilize the cyst and decrease the chance of anaphylaxis, and the tension in the cyst wall during the operation is used to reduce the risk of leakage and is maintained at least one month after surgery to prevent recurrence [11]. The scope of surgery depends on the location of the cyst, the age of the patient, the parity, and the degree of involvement of the reproductive organs. If a postmenopausal woman has multiple cysts in the reproductive organs, a total hysterectomy and bilateral salpingo-oophorectomy are required to remove the cysts. However, for young women, no matter how many cysts are present, every effort should be made to protect reproductive function [7]. Nowadays, intraoperative hypertonic saline or 0.5% silver nitrate solutions is used by most surgeons before opening the cavities to kill the daughter cysts to avoid propagation or anaphylactic reaction, which is safe and effective [11]. In addition, special attention should be paid to avoid cyst fluid overflow, which will cause allergic and inflammatory reactions in the fetus. 100mg hydrocortisone is usually used to prevent anaphylaxis caused by cystic fluid overflow. Secondary peritoneal echinococcosis is one of the important complications of cyst hydatid disease caused by the rupture of the original organ hydatid cysts, which may occur spontaneously, iatrogenically or by trauma, resulting in the reproductive elements initiating the formation of new cysts in the peritoneal and pelvic cavity [12]. According to reports, patients undergoing surgery has a 95% survival rate and a 2% recurrence rate, and the treatment only with albendazole is successful in a maximum of 40 % of cases [13]. In the present case, we noticed the frequent recurrences of the disease. Therefore, a total hysterectomy and bilateral salpingo-ophorectomy with complete removal of the cysts was performed by MDT surgical treatment combined with full course and full does albendazole treatment before and after surgery to reach successful consequence.

4. Conclusion

Although echinococcosis is mainly located in the liver, it can spread to many organs throughout the body, which require physicians take pelvic echinococcosis into consideration in differential diagnosis of pelvic masses. It is mainly treated by surgery, supplemented by drug therapy, to completely eliminate echinococcosis, reduce complications and improve survival rate and quality of life. It is important to perform a comprehensive and systemic evaluation before surgery to understand the relationship with the surrounding vital organs and exclude possible lesions involved. MDT treatment is also recommended to reduce collateral damages of multivisceral echinococcosis.

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