CASE REPORTS

A primary pelvic hydatid cyst causing primary infertility: management of this rare phenomenon and a brief review of literature

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Introduction

Primary pelvic hydatid cysts are a rare entity. According to the natural life cycle of the parasite, it should pass through the filters of liver and lung to reach anywhere else in the body. Intra-abdominal extra-hepatic hydatids are usually secondary to rupture of a primary hydatid cyst. For a pelvic hydatid to be considered a primary hydatid, there must be no evidence of hepatic or other primary sources [1]. Pelvic hydatid cysts can present non-specifically with pressure effects on adjacent organs. The bladder, rectum and fallopian tubes are often involved in compression. In extreme cases they can cause severe adjacent organ obstruction as well. We report a case of primary pelvic hydatid cyst detected in a 30-year-old female and the management of the condition.

Case Report

A 32 year old female presented with complaints of lower abdomen pain for the past 6 months. She has been married for 5 years and had long-standing primary infertility. Her menstrual cycles were regular and there was no history of pet exposure during her childhood. On examination there was no mass palpable and per rectal examination was normal, but per vaginal examination revealed a tender right sided adnexal mass.

Ultrasonography of the abdomen revealed a well-defined 7 x 8 cm multi-loculated cystic lesion in the pelvic region suggestive of a hydatid cyst. A CECT abdomen was done to confirm the diagnosis and to rule out any other disease sites (Figure 1). Liver function tests were normal, hydatid serology was negative and ovarian tumor markers were negative. A diagnosis of primary pelvic hydatid cyst was made and she underwent a laparoscopic hydatid cystectomy followed by omentoplasty and chromotubation for checking patency of the fallopian tubes.

Intra-operative findings revealed a large 8 x 11 cm hydatid cyst located posteriorly to the right ovary & fallopian tube

with dense adhesions. The liver and other organs were free of cysts. After packing the abdomen with 10% hypertonic saline gauzes a Palanivelu hydatid trocar system 2 was introduced into the cyst cavity directly under vision and cyst contents were evacuated. Repeated hypertonic saline wash was done. The superior portion of the cyst, which was not adherent to the ovaries, was dissected out. The remaining cavity dead space was obliterated with omentum. No cyst content spillage occurred during the procedure.

Figure 1. CECT abdomen showing hydatid cyst in pelvis.

Figure 2. Intra-operative view of hydatid cyst.

Post-excision chromotubation was done to check tube patency. Both the tubes were patent with dye spillage noted.

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The post-operative period was uneventful and patient was discharged on the 4th day with albendazole medication.

After 4 months she conceived and was on regular follow up with the obstetrician during her pregnancy.

Discussion

Atypical presentation of hydatid disease in the pelvis is not well explored in literature. According to a large case series of 540 cases by Prousalidis et al [3], only 5% of the patients had peritoneal disease. The most frequent presenting symptom in their series was abdominal pain secondary to the pressure effects. There are no specific symptoms of pelvic hydatid disease and the symptoms are determined by the size of the cyst [4]. These range from vague lower abdominal pain, swelling, pressure symptoms, menstrual irregularities to even infertility according to the case series by Rahman et al [4].

A few case reports exist regarding secondary pelvic hydatid cysts, but very few reports are present in the literature regarding a true primary pelvic hydatid disease. Of these few primary cysts, all of them presented with abdomen pain, urinary obstruction or asymptomatic disease [5]. So far there is no case report in the literature of a primary pelvic hydatid causing infertility in a patient. Although it cannot be said with 100% certainty that our patient's infertility was due to the cyst per se, we are convinced that the pelvic hydatid cyst could have been one of the causative factors for her infertility owing to the pressure effect of the large cyst on the fallopian tubes.

Diagnosis usually involves serology and imaging with ultrasound scanning being the preferred first investigation of choice. However, CECT offers important details regarding the site, number and relationship to adjacent vital structures.

The surgical treatment of hydatid disease can be broadly classified as radical or conservative. Radical procedures include pericystectomy, hepatic resection, and lobectomy. Conservative procedures include marsupialization; a simple cystectomy, omentoplasty or capitonnage [6-8]. There is an ongoing debate between radical approaches and conservative surgical approaches. In the series by Prousalidis et al, a partial pericystectomy was performed in most cases of the peritoneal

hydatid disease and the residual cavity was packed with omentum [3]. Outcome of this technique was good and there was no significant postoperative mortality or morbidity.

We adopted this same technique in our patient also with good results. Minimal access surgery is a safe & effective option for the management of selected patients with hydatid cysts in various locations.

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Key Points:

- Hydatid diseases are zoonotic diseases, which are common in the Asian countries. Extra hepatic sites of presentations and its management are not widely described in the literature.
- Pelvic hydatid cysts can be a rare yet reversible cause of infertility and unlike the other causes of infertility, these cysts are easily manageable surgically.
- Accurate pre-operative imaging and meticulous handling of the cyst intra operatively to avoid spillage are some important aspects to bare in mind when managing hydatid cysts.