CASE REPORT

AN UNUSUAL CASE OF CO-EXISTENT PRIMARY PELVIC AND INTRAMUSCULAR HYDATID CYST
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ABSTRACT: CONTEXT: Hydatid disease is a cyclo-zoonotic parasitic infection caused by Echinococcus granulosus. This disease is usually found in liver and lungs but no organ of body is immune. Location at unusual sites in the body can have atypical presentations and can pose a diagnostic challenge. A high index of suspicion, radiological investigations as well as histopathological examination is necessary in establishing the diagnosis of hydatid disease at unusual sites in the body.

SETTINGS AND DESIGN: We present a case of unusual extrahepatic occurrence of pelvic hydatid cyst with intramuscular extension to the rectus abdominis muscle and our experience with clinical presentation and management of hydatid disease. The case was treated successfully by surgical and medical methods.

CONCLUSION: Unusual presentations of hydatid disease can be successfully treated.

KEYWORDS: Hydatid disease, unusual, pelvic hydatid cyst, intramuscular extension.

INTRODUCTION: Hydatid disease has been known since the time of Hippocrates and it still represents a major health problem in endemic regions.¹ Echinococcosis is endemic in the Mediterranean countries. Hydatid cyst is the larval form of cestode tapeworm Echinococcus granulosus.² Hydatid disease is endemic in India, as well as other parts of the world, including Middle East, Africa, South America, New Zealand, Australia, Turkey and Southern Europe. Infestation by hydatid disease in humans most commonly occurs in the liver followed by the lung; the two organs can be affected simultaneously in about 5-13% of cases. It may present in an unusual manner in these usual sites.

The other rare sites reported to be involved by hydatid cyst are peritoneal cavity, spleen (5.1%), pancreas, thyroid, breast, gallbladder, thigh, kidney, brain, supraclavicular region, pericardium, diaphragm and pleural cavity.² Even though hydatid cysts can occur in any organ,³ it is very rare to see the disease in the sites reported in this article. Musculoskeletal hydatidosis is very rare and represents 0.5% - 4.7% of all cases of Echinococcosis.⁴

We present a rare case of extrahepatic occurrence of hydatid cyst in the pelvis with secondary musculoskeletal extension to the rectus abdominis muscle.

CASE PRESENTATION: A 26 year old female, p₁+₀, presented in our department with lower abdominal pain & swelling for two and half years which was gradually increasing for last six months. There was no history of rapid increase in size of the swelling. Patient complained of occasional dull aching pain over the right upper abdomen for last two years. There was a history of jaundice 15-20 days back before the admission in that hospital. She underwent total abdominal hysterectomy 3 years ago. She used to have beef for a long time.
General survey and systemic examination were essentially normal. On local examination, a firm mass was felt in hypogastric region of about 6×7 cm in dimension, and non-tender. On per vaginal examination, a 6×7 cm firm to cystic mass was felt through the anterior fornix and extended upto the right iliac fossa. No other mass was palpable. No free fluid was present in abdomen.

**Investigations:** USG showed the following feature: the Right adnexal area showed a large oval cystic mass with multiple septae suggestive of right sided tubo ovarian mass, measuring 127mm×48mm.

**Contrast enhanced CT scan of the whole abdomen showed the following features:** Liver – was normal in size, shape and echotexture. Multiple cystic space occupying lesions with septations & enhancing smooth walls were noted in the pelvis. Content in one of the cyst showed an attenuation of 40 hu & largest cyst measured 7.3×6.4 cm, which was suggestive of an ovarian origin. Liver function test was within normal limits. Serum CA-125 value was 4.40 U/ml.

Laparotomy was done under general anesthesia. After giving a suprapubic low transverse incision, abdomen was opened in layers. Right sided adnexal mass was approximately 15×10 cm whereas left sided adnexal mass was 10×10 cm. Both of them extended retroperitonially and sat over the great vessels. Gross adhesions were found between multiloculated cystic mass with that of the bladder and sigmoid colon which were separated by blunt and sharp dissection. Cavity wall was obliterated by deep stitches. A 5×5 cm cyst also found in left sided rectus abdominis muscle which was removed in similar fashion.

The images of the cysts during and after surgery have been pictorially shown in figures 1, 2 and 3. Abdominal wash was given with hypertonic solution. Peri and post-operative period was uneventful. Sutures were removed on the 7th post-operative day.

**Histopathology:** Both the adnexal as well as the rectus sheath masses were hydatid cysts.

Treatment post-surgery was by medical therapy with Albendazole [10 mg/kg BW for 3 month with 2 weeks gap between two cycle] to wipe out the rest of the scolices.

**DISCUSSION:** Hydatid disease or echinococcosis is a parasitic disease caused by infection with larva (metacestode) of the cestode echinococcus. Four species of the genus echinococcus are known to cause infection in humans: echinococcus granulosus (cystic hydatid disease), echinococcus multilocularis (alveolar hydatid disease), echinococcus vogeli, and echinococcus oligarthus (both causing polycystic hydatid disease)\(^5\). Echinococcus granulosus requires two hosts. Humans become accidental intermediate hosts. The most common site involved is the liver (59–75%), followed in frequency by lung (27%), kidney (3%), bone (1–4%), and brain (1-2%). Other sites such as the heart, spleen, pancreas, omentum, ovaries, parametrium, pelvis, thyroid, orbit, or retroperitoneum, and muscles are very rarely affected.\(^6\)

When the cyst is present in the rare site such as in musculoskeletal tissues, the suspicion of hydatid cyst in unlikely. This is further confusing when the patient does not have any primary hydatid cyst in lung or liver.

Peritoneal hydatid cyst, either primary or secondary, represents an uncommon but significant manifestation of the disease (approximately 13%). Intraperitoneal hydatid cysts are usually secondary to the rupture (spontaneous or accidental at surgery) of a primary hepatic, splenic, or
mesenteric cyst. A solitary cyst in the pelvic cavity can be considered primary only when no other cysts are present. In such a case, the hydatid embryo gains access to the pelvis by hematogenous or lymphatic route. Pelvic hydatid cysts usually present as a nonspecific mass with pressure effects on adjacent organs such as the rectum and urinary bladder.

Rarely, they can cause obstructed labor, obstructive uropathy, and renal failure. Sometimes, they can rupture spontaneously. Serology and imaging are the main tools for establishing diagnosis. Ultrasound is the preferred first-line imaging, but CECT (Contrast Enhanced CT) gives more precise information regarding the morphology (size, location, neighborhood, and number) of the cyst. Drug treatment with albendazole has been found to be successful in a proportion of cases, but drug therapy is generally not used as the primary treatment except in cases where the patient is not fit for surgery or the cyst size is smaller or deeply located. Surgery is the most effective treatment.

Combination of preoperative albendazole therapy, surgery, and postoperative albendazole therapy is a useful regime. Albendazole suppresses the development of hydatid cysts following intraperitoneal inoculation of protoscolices. En bloc resection without inducing rupture and spreading the daughter cyst is recommended treatment strategy and accepted to be curative for intramuscular hydatid disease. Partial cystectomy, however, is another commonly practiced modality of surgery where the surrounding adhesions or the removal of ectocyst is considered to do more harm than good.

The treatment of choice in musculoskeletal hydatid disease is surgical excision (pericystectomy), potentially combined with antihelminthic medication. Percutaneous aspiration, infusion of scolicidal agents and reaspiration (PAIR), under imaging (ultrasound or CT) guidance can be used as alternative to surgery in inoperable cases. Aspiration of fluid is safe, simple and an effective means to reach a working diagnosis.

CONCLUSION: Pelvic echinococcosis is rare, with an incidence of 0.2-2.25%. Pelvic hydatid cysts may have varied presentation. It has to be differentiated from mesenteric cyst. Ultrasonography and computed tomography are both excellent imaging modalities for the detection of hydatid cysts. The treatment of choice for pelvic hydatid cyst is principally a careful and complete surgical excision.

REFERENCES:


Fig. 1: Photograph of the pelvic hydatid cyst being operated

Fig. 2: Photograph of the intramuscular hydatid cyst being operated
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Fig. 3: Photograph of the cyst wall of the pelvic and musculoskeletal hydatid cysts after laparotomy

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