International Surgery Journal
Shukla S et al. Int Surg J. 2020 Jan;7(1):287-291
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: http://dx.doi.org/10.18203/2349-2902.isj20195987

Case Report

Parametrial hydatid cyst with cystovesical fistula: a rare presentation

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Received: 20 October 2019
Revised: 23 November 2019
Accepted: 29 November 2019

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ABSTRACT

Hydatid disease is a parasitic infestation by a tapeworm of the genus *Echinococcus*. Theoretically, echinococcosis can involve any organ. The liver is the most common organ involved, followed by the lungs. However, uterus and and adnexa have an incidence of 0.5-1%. Till date, three cases have been reported in which a parametrial hydatid cyst formed a bladder fistula. A 57 year old female was admitted with complaint of burning micturition along with white coloured urine for the last 15-20 days with no other significant history. On examination and evaluation, it was suspected to a neoplastic pelvic mass causing gross right sided hydronephrosis. All biochemical markers were within normal limits. The patient was explored surgically on which a thick-walled lesion was found in right parametrium. The cavity of the lesion was filled with daughter hydatid cyst, along with communication into the bladder via a fistula. Open cystectomy was done after evacuation of daughter cysts and irrigation with scolicidal agents. The fistula was repaired. The patient recovered uneventfully and was doing well on follow-up. The incidence of hydatid cysts in the female reproductive system is very low and constitutes less than 0.5% of all hydatid cysts. However, rare cases have been reported, thus the clinician must be aware of this rare disease and should take precautions while operating, as any spillage may lead to anaphylactic shock.

Keywords: Hydatid cyst, Parametrium, Bladder fistula, Cystovesical fistula

INTRODUCTION

Hydatid disease is a parasitic infestation by a tapeworm of the genus *Echinococcus*. Of the four known species of *Echinococcus*, three are of medical importance in humans. These are *E. granulosus*, causing cystic echinococcosis (CE); *E. multilocularis*, causing alveolar echinococcosis (AE); and *E. vogeli*. The endemic areas are the Mediterranean countries, the Middle East, the southern part of South America, Iceland, Australia, New Zealand, and southern parts of Africa; the latter five regions are intensive endemic areas. Central Asia, particularly China, is also an endemic area. In India, hydatid disease is common in most of the states with preponderance in Andhra Pradesh and Tamil Nadu.[1]

It is one of the most widespread zoonotic diseases in humans in both developing and developed countries.[2] Humans act as an accidental intermediate host. The intermediate host ingests the eggs, which hatch into metacestodes, which infest the liver, lungs, muscles, and other organs of the intermediate host.[3]

Following is a case report of a rare presentation of parametrial hydatid cyst with bladder fistula.

CASE REPORT

The patient, a 57-year-old female was admitted with complaint of burning micturition along with white coloured urine for the last 15-20 days with no other

significant history. She had previous history of a pair procedure done 5 years back for a hepatic hydatid cyst. There were no significant findings on physical examination. The USG was suggestive of large multicystic pelvic cavity mass lesion suspicious of ovarian neoplasm along with gross right hydronephrosis. CECT Abdomen was suggestive of large well-defined thick-walled lesion in pelvic cavity showing multiple variable sized daughter cysts within and calcification on anterior wall. All biochemical markers like complete blood count, total leucocyte count, differential leucocyte count, liver function test and renal function test were within normal limits.

The patient was explored surgically. There a thick-walled lesion was found in right parametrium. The cavity of the lesion was filled with daughter hydatid cyst, along with communication into the bladder via a fistula. Open cyst evacuation and irrigation with 10% povidone-iodine solution and 10% hypertonic saline was done. Thereafter the fistula was repaired.

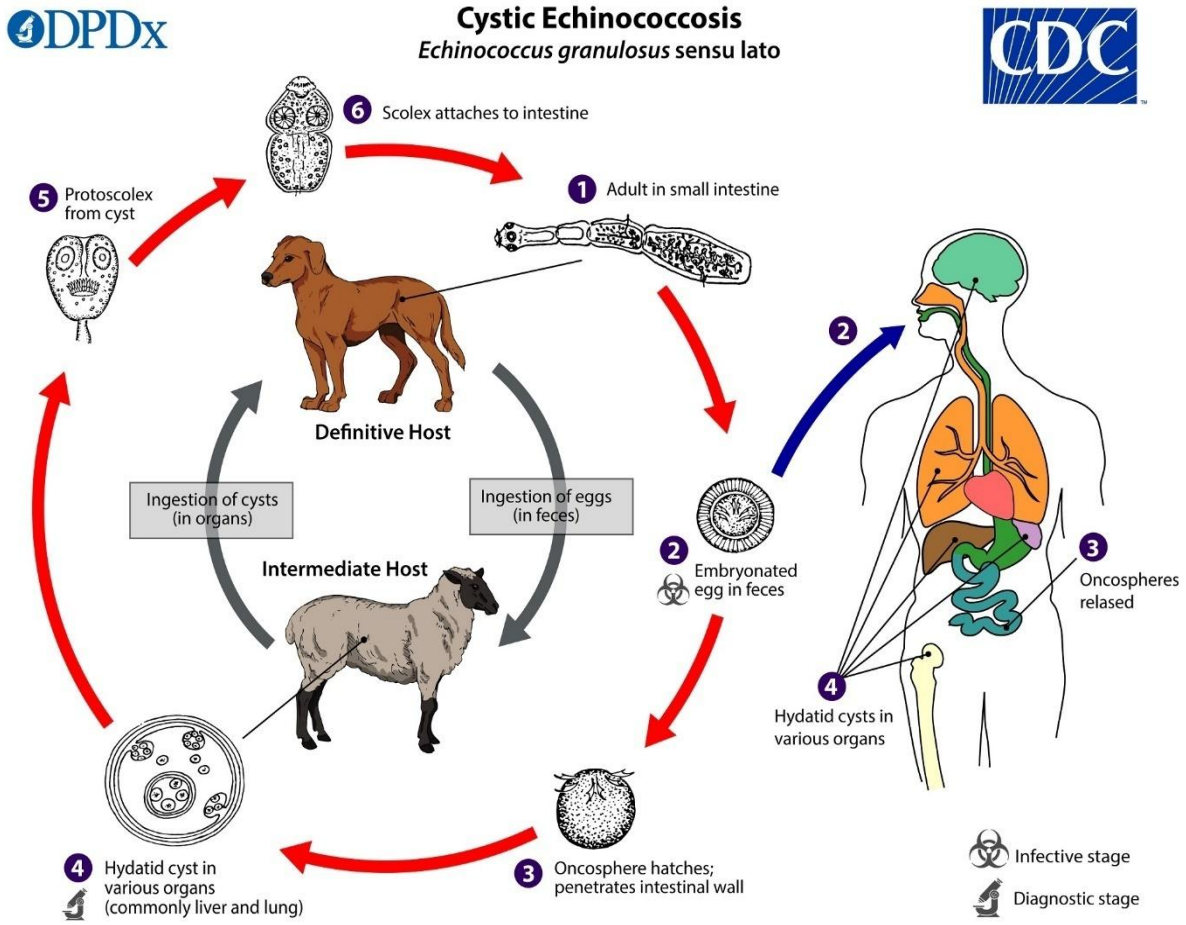

Figure 1: Lifecycle of *Echinococcus granulosus*.[3]

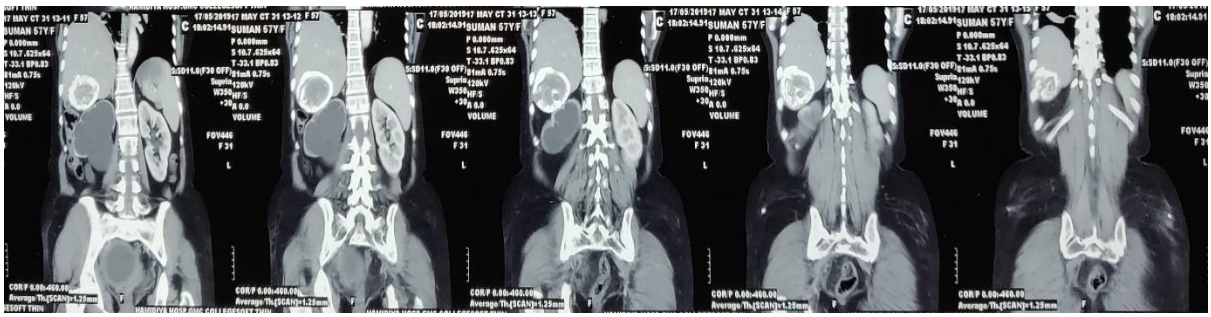

Figure 2: CECT abdomen of the patient -coronal sections.

Figure 3: CECT abdomen of the patient-axial sections showing old calcified lesion in the liver.

Figure 4: CECT abdomen of the patient-axial sections showing the cystic lesion with daughter cysts within it.

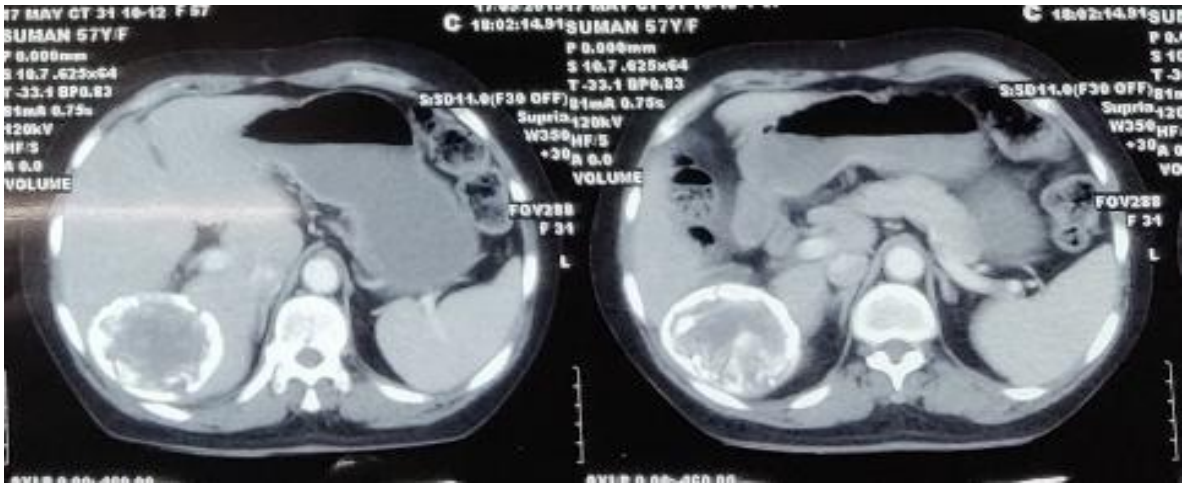

Figure 5: CECT abdomen of the patient-axial sections showing old calcified hepatic lesion.

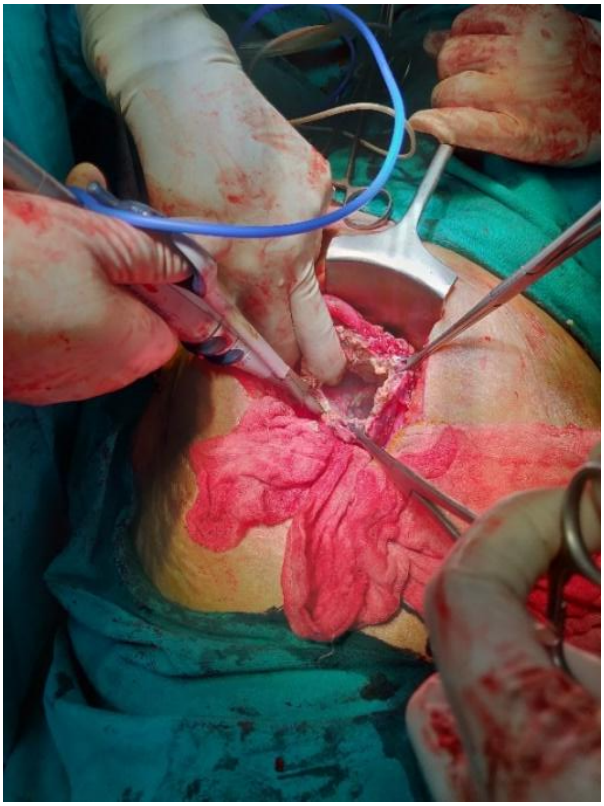

Figure 6: Intraoperative image of open cyst evacuation-the cyst is opened and its contents are visible.

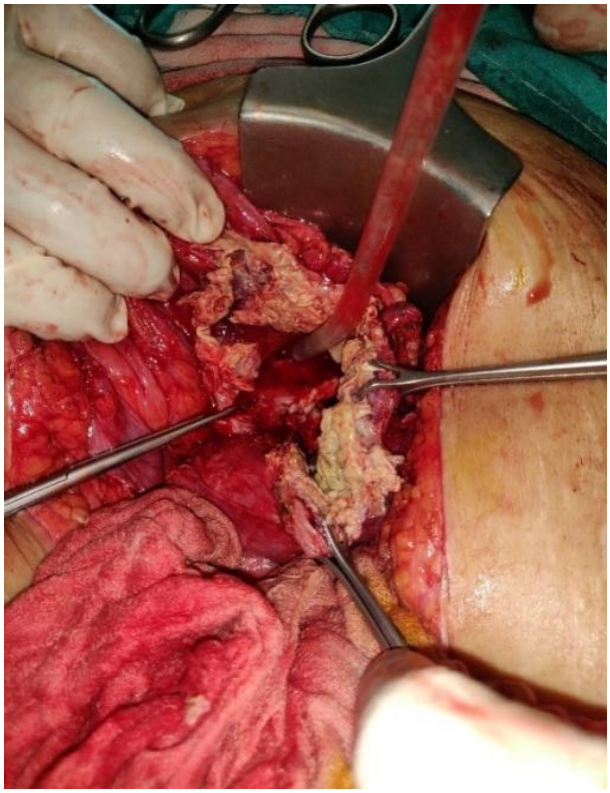

Figure 7: Intraoperative image showing evacuation of the cyst.

Figure 8: Post operative image of the retrieved daughter cysts.

DISCUSSION

Hydatid disease is a parasitic infestation by a tapeworm of the genus *Echinococcus*. Of the four known species of *Echinococcus*, three are of medical importance in humans. These are *E granulosus*, causing CE; *E multilocularis*, causing AE; and *E. vogeli*. Their life cycle involves only two hosts, one definitive and the other intermediate. Humans act as an accidental intermediate host. The life cycle has three developmental stages, the adult tapeworm in the definitive host, eggs in the environment, and) the metacestode in the intermediate host. Metacestodes are ingested by the definitive host. The metacestodes mature into the tapeworm in the definitive host and, in turn, release eggs into the environment. The intermediate host ingests the eggs, which hatch into metacestodes, which infest the liver, lungs, muscles, and other organs of the intermediate host.[4]

The endemic areas are the Mediterranean countries, the Middle East, the southern part of South America, Iceland, Australia, New Zealand, and southern parts of Africa; the latter five regions are intensive endemic areas. Central Asia, particularly China, is also an endemic area. In India, hydatid disease is common in most of the states with preponderance in Andhra Pradesh and Tamil Nadu.[1]

No sexual predilection is recognized for hydatid cysts.

Because of the restricted geographic distribution of the echinococcal worms, persons of certain races are affected more commonly than others; however, the parasite has the capability of infecting persons of all races equally.

The cysts grow slowly, and a cyst is rarely diagnosed during childhood or adolescence unless the brain is affected. CE is a disease of younger adults, with an average age at diagnosis of 30-40 years, whereas AE is a disease of older adults, with an average age at diagnosis of older than 50 years.

Theoretically, echinococcosis can involve any organ. The liver is the most common organ involved, followed by the lungs. These 2 organs account for 90% of cases of echinococcosis. However, uterus and and adnexa have an incidence of 0.5-1%.[5,6]

Till date, three cases have been reported in which a parametrial hydatid cyst formed a bladder fistula.[7,8]

Imaging studies

Plain films

In CE, findings from plain films of the chest, abdomen, or any other involved site are, at best, nonspecific and mostly nonrevealing. A thin rim of calcification delineating a cyst is suggestive of an echinococcal cyst.

In AE, results from plain films may be normal.

Ultrasonography

Ultrasonography helps in the diagnosis of hydatid cysts when the daughter cysts and hydatid sand are demonstrated. The accuracy of ultrasound evaluations remains operator-dependent.

Computed tomography scanning

Computed tomography (CT) scanning has an accuracy of 98% and the sensitivity to demonstrate the daughter cysts. It is the best test for the differentiation of hydatid from amebic and pyogenic cysts in the liver.

In AE, the CT scan findings are sometimes indisting-uishable from those of hepatocellular carcinoma.

Magnetic resonance imaging (MRI)

Images show the cysts adequately, but MRI offers no real advantage over CT scanning.

Surgical care

Choice of surgical technique: Radical surgery (total pericystectomy or partial affected organ resection, if possible), conservative surgery (open cystectomy), or simple tube drainage of infected and communicating cysts for surgical options. The more radical the procedure, the lower the risk of relapses but the higher the risk of complications. Patient care must be individualized accordingly.

Laparoscopic approach has gained more acceptance and popularity in recent years.

CONCLUSION

Incidence of hydatid cysts in the female reproductive system is very low and constitutes less than 0.5% of all hydatid cysts. However, rare cases have been reported, thus the clinician must be aware of this rare disease and should take precautions while operating, as any spillage may lead to anaphylactic shock. We recommend that surgeons, gynecologists, radiologists, and histopatho-logists should suspect hydatid cyst whenever a cystic pelvic mass is found.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Shukla S, Khare VS, Koshariya M, Lal V, Parmar B, Tiwari S, et al. Parametrial hydatid cyst with cystovesical fistula: a rare presentation. Int Surg J 2020;7:287-91.