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Primary Pelvic Hydatid Cyst of Broad Ligament Mimicking Ovarian Cyst: A Rare Case Report.

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Abstract :
Hydatid disease is an anthropozoonosis. It is caused by Echinococcus granulosus in tropical countries. Humans are the accidental intermediate hosts of Echinococcus. It affects the liver and lung most commonly, but may rarely affect the fallopian tube, broad ligament and other structures. We report and discuss a case of primary pelvic hydatid cyst of broad ligament in a woman, who was suffering from primary sub fertility for last 15 years, presented with lower abdominal pain, clinically and sonographically suspicious of ovarian cyst preoperatively. Investigations failed to identify the nature of the disease until surgical exploration and microscopic studies confirmed the diagnosis. The patient responded well to surgical excision followed by albendazole administration. Female genital tract hydatidosis is a rare entity and in most cases the involvement is secondary. Primary hydatid disease of female genital tract is even very rarer and generates considerable diagnostic difficulty. However, diagnosis of pelvic hydatid cyst in females is possible only after operation. The gynecologists should be aware of possibility of primary hydatid cyst of the pelvic cavity and should be considered in the differential diagnosis of cystic pelvic masses, especially in areas where the disease is endemic, so that the patient can be managed accordingly. The objective of this presentation is to highlight a primary pelvic hydatid cyst of broad ligament that presented as an ovarian cyst.

Keywords : *Broad ligament hydatid cyst, Echinococcus, Primary pelvic hydatid cyst.*

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I. Introduction :

Hydatid disease is an anthropozoonosis. It is caused by the larval stage of *Echinococcus granulosus* and *Echinococcus multilocularis*, a parasite of the class Cestoda (tapeworm) and family Taeniidae. The disease is endemic in sheep and cattle grazing countries like India, Australia, Middle East, Africa and South America **(1,2)**. The life cycle of *Echinococcus* requires a definitive host, which is often a dog, and an intermediate host, which is commonly sheep. Humans become the accidental intermediate hosts when they become infected after ingesting ova passed in dog feces. Embryos migrate through the intestinal mucosa and enter the venules or lymphatic vessels. The liver, lung, and spleen are the most commonly affected organs. Hydatid disease of the pelvis is very rare and therefore, difficult to diagnose and treat **(3).** Bickers (1970), after reviewing 532 cases of hydatid disease from an endemic area over a 20-year period, recorded 12 instances where hydatid cysts were present in the pelvis, only 2 of which were in the broad ligament, so an incidence of 0.37% occurring in the broad ligament **(4).**

Pelvic hydatid cyst can present with vague abdominal pain due to irritation and swelling; menstrual irregularities, infertility, and pressure symptoms due to mass effect on the urinary bladder, ureters, rectum and vessels (some time ruptures) **(5,6,7)**.

We report and discuss a case of primary pelvic hydatid cyst of broad ligament in a woman, who was suffering from primary sub fertility for last 15 years, presented with lower abdominal pain, clinically and sonographically suspicious of ovarian cyst preoperatively. The aim of this case report is to highlight a rare presentation of primary pelvic hydatid disease located in the broad ligament that presented as an ovarian cyst.

II. Case Report :

A 35-years-old nulligravida lady presented to our Gynaecology OPD of Bankura Sammilani Medical College and Hospital, Bankura, West Bengal, India with the chief complaint of lower abdominal pain for last 4-5 months. She was also suffering from primary sub fertility for last 15 years. There was no associated fever or loss of appetite. Her bladder and bowel habits were normal. Her menstrual cycle was regular.

There was no history of recent travel. No history of tuberculosis, asthma, allergy nor were there dogs or any pets where the patient lived. The patient denied having been on a farm.

On general examination, the lady was thin built, well-nourished, and mild anaemic. There was no lymphadenopathy. Her blood pressure (BP) was 110/70 mmHg, with pulse rate 80/min, temperature normal, with mild pallor. On systemic examination, respiratory, cardiovascular and neurological systems were normal.

On per abdominal examination, the abdomen was soft and lax. A cystic mass of 20 weeks size with smooth surface was palpable in the left lower abdomen with restricted mobility. The mass was non tender. There was no hepatosplenomegaly or ascites. On per vaginal examination, uterus was normal size with restricted mobility. Cervix was healthy, right fornix free, left fornix full with a fixed, non tender cystic mass about 15×10cm.size, not separately felt from the uterus.

Urine for pregnancy test was negative. Routine blood and urine examination, chest X-ray, E.C.G revealed no abnormality. Serum CA-125, carcino embryonic antigen (CEA) were within normal limits. Ultrasonography (USG) of whole abdomen was suggested and it revealed a multicystic SOL of 10×9 cm. at left adnexa. The other abdominal organs were normal and there was no free fluid in the abdomen.

A provisional diagnosis of left sided ovarian cyst was made. The patient was electively prepared for exploratory laparotomy and proceed. Under spinal anesthesia, the abdomen was opened through left paramedian incision . After opening the abdomen, it was found that left sided broad ligament was diffusely enlarged and thickened , occupied by a large firm to cystic mass of about 15×10cm. size displacing the uterus to the right with the left fallopian tube stretched over the mass. Both ovaries were normal. No ascites found. During dissection, the cyst was unintentionally ruptured and multiple daughter cysts came out with spillage of intracystic clear fluid. Immediate suction of the fluid and complete evacuation of the daughter cysts from the cavity were done after meticulous packing of the operating field with mops. Only partial removal of the cyst was possible as a small part of the cyst wall was adherent to the rectum and uterus. It was not possible to dissect the cyst wall completely and so marsupialization was performed on the remaining cyst wall to avoid injury to the rectum and uterus. No other intra abdominal pathology was found. The liver was explored and no lesions were found. Thorough peritoneal toileting and irrigation of the cyst cavity was done with hypertonic saline many times and a suction drain was left in the pouch of Douglas. The abdomen was closed in layers after taking count of the instruments and the mops. Specimen of the cyst wall with daughter cysts were sent for histopathological examination. Gross features the specimen were consistent with hydatid cyst; the cyst wall was white and there were multiple small thin wall daughter cysts **(Figure 1).**

Figure 1: Showing white cyst wall with multiple small thin wall daughter cysts.

The patient was shifted to HDU for monitoring and the risk for anaphylactic shock during postoperative period. Injection Hydrocortisone and injection Phenergan were given during intra/postoperative period prophylactically for prevention of anaphylaxis. General Medicine referral was given. They advised Tab

Albendazole 400mg twice daily for 1 month and further review with CT Scan of whole abdomen and thorax. Postoperative period was uneventful. The liver function and renal function test were monitored twice weekly together with the complete blood count and coagulation profile. On 10th postoperative day CT scan was done. MDCT Scan of whole abdomen (Oral & I.V. Contrast) showed only hyperdense sludge in the gallbladder lumen. HRCT Scan of thorax (Plain) showed patchy areas of ground glass opacity with intervening lucency noted in bilateral lung segments. Patient was discharged in satisfactory condition after stitch removal on 11th postoperative day and was advised to attend General Medicine OPD, Chest Medicine OPD along with Gynaecology OPD. She was put on postoperative chemotherapy of albendazole and is doing well on follow-up.

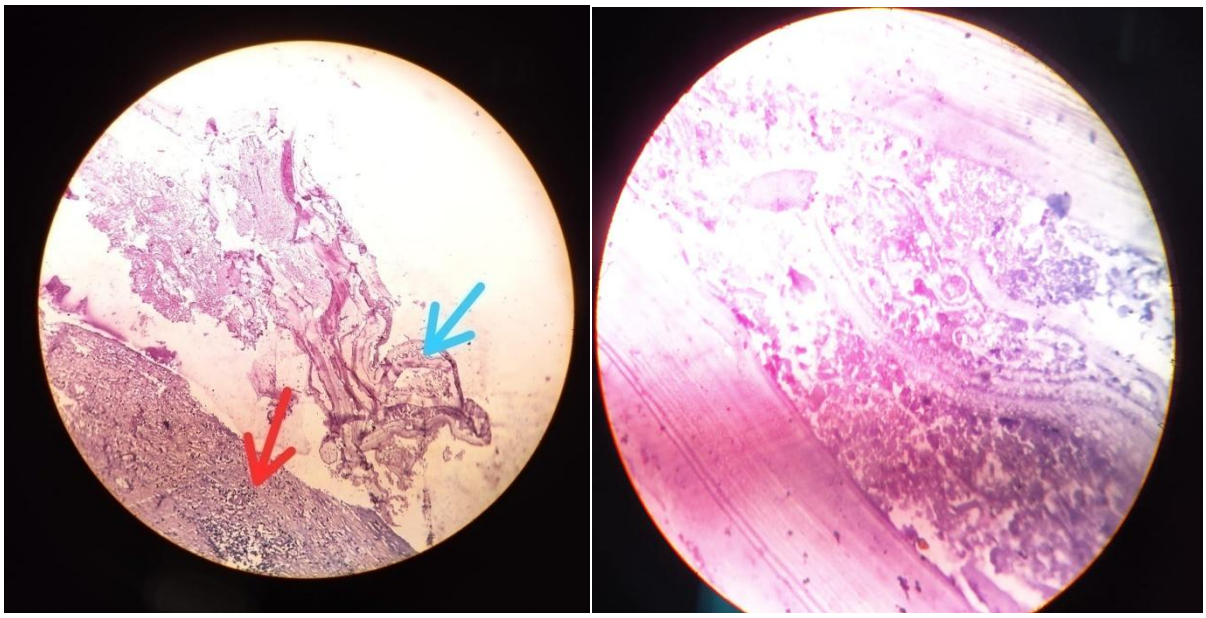

Figure 2: Microphotograph of the lesion reveals the laminated Hydatid Cyst layer (blue arrow) and the broad ligament tissue (red arrow).

Multiple sections of the lesion stained and showed the histopathological features of Hydatid Cyst. In microphotograph of the lesion, the blue arrow is showing the hydatid cyst wall. Laminated appearance indicates the middle layer of hydatid cyst wall. The fibrous parts indicate the outer pericyst layer. Red arrow is showing the broad ligament tissue with the features of inflammation **(Figure 2)**.

III. Discussion :

Hydatid cysts are characterized by indolent yet unremitting growth in the majority of patients, with the potential for cyst metastasis to the peritoneal surface and the lungs **(8).** Peritoneal hydatid cyst disease can be primary or secondary to hydatid cyst in the liver, spleen or any other abdominal organ; with the pelvis being affected due to spillage during primary surgery **(9)**. Primary peritoneal hydatidosis is rare and accounts for only 2% of all abdominal hydatid diseases **(10).** Involvement or occurrence of cyst in pelvis is rare and primary pelvic hydatid cyst is extremely rare **(11).** A solitary pelvic hydatid cyst is considered primary when no such other lesion is found elsewhere in the body as in our case. In such cases, the invasion is considered to be by the haematogenous or lymphatic route.

One such exceptional case of primary pelvic hydatid cyst of broad ligament is being described hereby as the condition is rare and clinically difficult to identify because the unusual site for the disease. Cystic lesion in the female pelvis is commonly encountered as seen in our case **(12)**. The symptoms of the disease in this case are not so specific, they simulate those of ovarian cysts except long-standing primary sub fertility.

It is usually diagnosed intra operatively or postoperatively **(13)**. The pre-operative diagnosis of hydatid disease may be possible by radiographs, ultrasonography and CT scan in the women with high clinical suspicion of hydatid cyst. The plain radiograph may show calcification in the cyst wall. Ultrasonography and CT scan may demonstrate the features like multilocular appearance, a fluid level from hydatid sand and ultrasonic 'water lily sign'. In this study case, ultrasonography revealed a multicystic adnexal SOL. Casoni's test is only 63.8% sensitive and 47% specific to hydatid disease and has been superseded by more sensitive, specific and safer serological tests.

Preoperative anthelmintic (albendazole) therapy followed by the surgery is the preferred treatment for hydatid cyst. En bloc excision of the cyst without rupture is accepted as curative. Partial cystectomy, however, is

the other modality where surrounding adhesions cause problems or removal of the ectocyst is dangerous **(14,15,16)**. Only partial cystectomy followed by marsupialization on the remaining cyst wall was done in this index case as a small part of the cyst wall was adherent to the rectum and uterus.

IV. Conclusion :

The occurrence of primary pelvic hydatid cyst of broad ligament is rare with a reported incidence of less than one percent and can pose considerable diagnostic dilemma in female patients. A high degree of clinical suspicion is the most important factor in establishing preoperative diagnosis. The other important consideration is the accidental rupture of hydatid cyst during surgery which may be life threatening, therefore preoperative diagnosis of this rare lesion is very important. Awareness regarding *Echinococcosis* as a case of cystic pelvic lesion, especially in endemic zones, will avoid the diagnostic problems and prevent the erroneous treatment, thereby avoid the most potentially serious complications.

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Conflicts of interest :
There are no conflicts of interest.

Ethical approval :
Not required.

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