Case Report

# Primary ovarian hydatid disease in the Kingdom of Saudi Arabia

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## ABSTRACT

Human hydatid disease is caused by *Echinococcus granulosus*. Its distribution is world wide and it affects mainly the liver, but other organs could be involved. Primary involvement of pelvic organs is very rare. This is a case report of primary ovarian hydatid disease in a postmenopausal woman, diagnosed postoperatively. Surgical excision was adequate. Ultrasonography, particularly high frequency trans-vaginal, computed tomography scan and, more recently, magnetic resonance imaging are more frequently used in the diagnosis of *Echinococcus* cyst. They appear more reliable than many of the old tests of varying sensitivities. Whereas, there are anecdotal reports of obstetric and gynecological manifestations of echinococcosis from some Middle Eastern and North African countries, this is the first of such report from the Kingdom of Saudi Arabia. It is unclear why there is a lack of information about this condition among Saudi women, even though socio-cultural attitude to female involvement in sheep farming and animal husbandry is similar to that in other Arabic and Islamic countries. We endorse the recommendation that every gynecologist, radiologist and histopathologist should maintain a high index of suspicion for hydatid cyst, whenever a septate cystic pelvic mass is found.

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Hydatid disease in humans is caused by the larval form of parasites of the genus *Echinococcus*.[1,2] The most common form is cystic hydatid disease resulting from *Echinococcus granulosus (E. granulosus)*. Humans become an accidental intermediate host when contaminated, unwashed vegetables are ingested or through contact (usually in childhood) with infected animals, which may carry ova on their fur or shed ova on to soil in which children play. When ingested, these ova follow the same route through the intestine in humans to form cysts such as in liver or lung. Growth is often slow, and it may take 5 to 20 years before hydatid cyst become symptomatic.

Human disease caused by *Echinococcus* species results from blood-borne invasion of the liver (50-70% of patients), lungs (20-30%) or other organs such as the brain-causing hydrocephalus and seizures, and bone resulting in pathological fracture. There had been some reports of hydatid disease affecting both male and female pelvic organs. However, involvement of the reproductive organs is rare.[3] We report here a case of hydatid disease of the ovary. As far as we know, this is the first report of hydatid disease of the ovary in the Kingdom of Saudi Arabia (KSA), a country where hydatidosis is very common.

**Case Report.** A 70-year-old grand multiparous woman was referred from a small peripheral hospital with history of abdominal swelling of 2 months duration. This had become constantly painful for about 2 weeks. She had been

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**Figure 1 -** Photomicrograph showing part of the wall of the hydatid cyst and degenerated daughter cysts (top right) with a protoscolex showing hooklets. Note abundant scattered granular debris (hydatid sand).

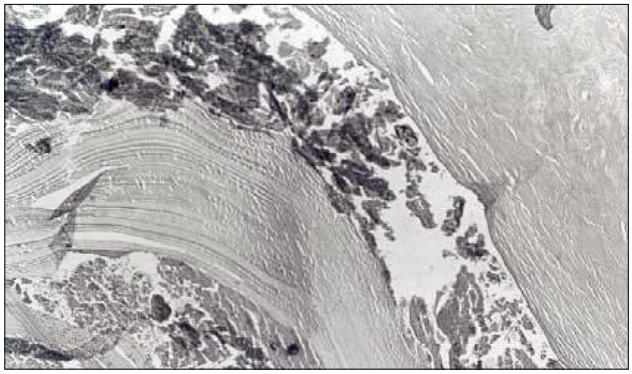

**Figure 2 -** Photomicrograph showing trilaminar cyst wall with degenerated brood capsules and protoscolices amidst necrotic debris. Note the residual ovarian tissue to the left.

menopausal for 20 years. Her only previous admission to any hospital was when she suffered transient ischemic attack 2 years earlier, but currently not on any medication.

Clinical examination revealed a mildly obese elderly woman. Her weight was 97.5kg and height 161cms. Her vital signs were normal. There was mild fullness and tenderness in the lower abdomen. A poorly defined mass arose from the pelvis and was compatible with a pregnancy of about 16 weeks duration. There was no ascites. Ultrasonography showed a right multi-lobulated mass measuring 114x119x67mm, which was suspected to be ovarian in origin. There were some solid areas. No free fluid in the pelvis or abdomen. The provisional diagnosis was complicated ovarian mass. She was admitted for preoperative work-up and laparotomy for total hysterectomy and bilateral salpingo-oophorectomy. The patient and family refused hysterectomy. Following admission, full cardiological assessment, including ECG and echocardiograph, did not reveal any cardiac abnormality.

Two days after admission, she developed acute exacerbation of her pain, emergency exploratory laparotomy had to be performed. At operation, through a midline sub-umbilical incision, moderate to severe flimsy peritoneal adhesions were present involving the uterus and its appendages, the small intestines and the omentum. The uterus, right ovary and right fallopian tube were atrophic. There was a huge left tubo-ovarian mass extending posteriorly into the pouch of Douglas (and following excision, it weighed 500g). Small multiple caseous-looking nodules were seen on the free border of the omentum. No obvious enlarged lymph nodes were seen or palpated. The liver surface was smooth. Bilateral salpingo-oophorectomy and omental biopsy were performed since consent for hysterectomy was refused. An intra-operative diagnosis of tuberculous tubo-ovarian abscess was made.

Postoperative recovery was un-eventful, apart from hyperglycemia requiring insulin injections, and later oral hypoglycemics. She was discharged home on the ninth postoperative day. At the outpatient follow-up clinic at 4 and 8 weeks postoperatively, she was asymptomatic and well. The wound had healed satisfactorily and no abnormality was detected. She defaulted from further follow-up visit.

The complete blood count including white blood count differentials, renal function tests, liver function tests, random blood sugar and urinalysis were normal. Blood group was O positive. Chest x-ray was normal but intravenous pyelogram, which is very useful in delineating the course of the ureters in pelvic masses, could not be carried out due to temporary mechanical fault. Computed tomography (CT) scan showed a multi-cystic pelvic-abdominal mass measuring 100x100x55mm. The outer cyst wall was smooth, while the internal wall showed some specks and calcification. There was no pelvic or para-aortic lymphadenopathy and no ascites.

Histopathology report revealed small fragment of ovarian tissue showing presence of fibro-collagenous layer (pericyst), an inner partially calcified layer (ectocyst), which appeared fused with innermost granular (endocyst) layer (**Figure 1**). Multiple brood capsules containing daughter cysts were present on the granular layer (**Figure 2**).

***Summary of histopathological report***. Hydatid cyst of the left ovary and chronic bilateral non-specific salpingitis.

**Discussion.** Of all cases of abdominal hydatidosis, 6.7% present with extra-hepatic lesions and only 0.7% were seen to have pelvic hydatid cysts.[2] It has been suggested that approximately 80% of all pelvic hydatid disease involve the reproductive organs, the ovary being the most frequent location. An exceptional case of bilateral

hydatid cysts of the fallopian tubes of a young girl living in endemic area had also been reported.[4] In these cases, symptoms and signs are usually referable to local compression of the genital organs, urinary tracts, vascular and bony structures. Primary involvement of pelvic organs by *E. granulosus* is extremely rare and is almost always secondary to rupture into the peritoneum of hepatic cyst or cysts of other abdominal organs.[1] However, primary pelvic echinococcosis has been described.[5] In such situations, the disease appears to be exclusively confined to the genital organs, which are considered to be the primary site of inoculation through hematogenous route.

In this patient, preoperative ultrasonography and CT scan of the abdomen were carried out to identify any probable metastases from this ovarian mass, should it turn out to be malignant. These investigations also excluded any hepatic or pulmonary lesions, the common sites of *Echinococcus* cyst. At laparotomy, as there was no other organ involvement, it is most probable that this was a primary hydatid infestation of the ovary.

Diagnosis of hydatid disease depends on tests, which have varying sensitivities and some are even poor indicators of active infection.[6] Mononuclear leukocytosis, eosinophilia, or high erythrocyte sedimentation rate may be absent in established disease situations, when the cyst has grown. In our patient, the total white cell count as well as monocyte and eosinophil counts were normal. Indirect hemagglutination test (IHAT) levels are said to be more reliable, especially for postoperative follow up. The most specific test, anti-*E. granulosus* immunoglobulin E antibody, is available only in very few countries where the disease is endemic.[7]

Ultrasonography (particularly high frequency trans-vaginal) and CT scan have been well-established techniques in the evaluation of hydatid disease and follow-up.[3,8] They appear to be the most helpful of all ancillary investigations. In ultrasonographic examination, benign cyst usually appears as simple echogenic structures with a high resistance index.[9] Peculiar onion-slice pattern is highly suggestive of the disease. Where there had been secondary calcium-folds formation and neo-vascularization of the pericyst, it may give the typical maize-like appearance also described in *Echinococcus* cyst.[10] In contrast, high neovascularization of a cyst both externally and internally and with low resistance pattern is diagnostic of ovarian malignancy.

The resolution of CT scan is excellent in the evaluation of hydatid cyst and it is better at detecting calcification in the cyst wall, which is visualized as thin dense rim of tissue.[11] It is said to be superior to ultrasound in precisely defining the extent of abdominal cyst and illustrating the abdominal structures after distortion caused by the cyst. The abdominal CT scan report in this patient could have raised our index of suspicion of a hydatid cyst of the ovary, especially as there were some specks and calcifications in the internal wall of the cyst. This could be attributed to the rare nature of, and our lack of experience in, this type of ovarian pathology.

More recently, magnetic resonance imaging signal characteristics have also been reported.[12] Undoubtedly, the experience of the radiologist particularly in an endemic area will promote a higher index of suspicion leading to the diagnosis. It has been suggested that in endemic areas, the possibility of hydatid disease should be considered in the differential diagnosis of any mass or growing tumor in the female pelvis.[1] Surgical removal is the usual treatment in women with pelvic hydatid disease.[13] Where surgical excision is incomplete or there had been spillage of cyst content at operation, oral mebendazole and albendazole have proved to be quite effective.[3,14]

Echinococcosis is endemic in many parts of the Middle East as well as other parts of the world including India, Africa, South America, New Zealand, Australia, Turkey and Southern Europe.[15] There are many anecdotal reports of pelvic hydatidosis in the female, emanating from hospitals in Libya, Tunisia, Turkey and Morocco.[1,4,15-17] Even in some of these cases, the disease has been associated with pregnancy and had been a cause of obstructed labor.[14] Apart from one report from Kuwait of ovarian hydatid disease presenting like ovarian carcinomatosis,[3] and another from Abha in KSA (not directly involving the pelvic organs)[13] there had been no other report from the Gulf region or the Middle East on female pelvic hydatid disease. It remains unclear why there is such a dearth of information about obstetric and gynecological manifestations of this disease in this country, though KSA shares with other Arab countries in the Middle East and North Africa similar socio-cultural attitude to female involvement in animal farming and pets. It is either such cases are missed or there may be a strong immunological factor among the female population (unlike the male population), which is protective. With such an excellent health care delivery and communication systems, it is unlikely that the diagnosis has been frequently missed, as histopathological services are within the reach of most secondary care institutions in the KSA. This is an area needing further study. This report should once again serve to raise the awareness of obstetricians and gynecologists, radiologists and pathologists working in endemic areas, as has been previously advocated.[1,8,12,13]

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