

Pelvic Hydatid Cyst: Three Cases with Suspected Adnexal Masses

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Abstract

Hydatid cysts are caused by *Echinococcus granulosus*. The usual organs in which the parasite lodges are the liver and lungs. One of the most peculiar locations for cyst formation is the ovary and most of these cases are secondary. It has been reported that some cases are formed iatrogenically. In our practice, we encountered three cases of ovarian hydatid cysts with a common symptom of abdominal pain. Imaging studies showed the presence of cystic lesions. Our first case was particularly interesting as the ultrasonographic characteristics were consistent with malignancy; however, surgical intervention showed that she had a hydatid cyst. Of note, this appeared to be a primary cyst because no other lesion was detected at the time. Our second case was diagnosed with infertility and under treatment prior to referral to our centre. Ultrasound (US) imaging showed the presence of a large cyst. The patient underwent laparoscopic cystectomy and the parasitic lesion was removed; however, she was subsequently diagnosed with urticaria and admitted to the intensive care unit (ICU). She made an uncomplicated recovery. Our third case was a pregnant woman with a previous diagnosis of a hydatid cyst. She underwent surgery for a suspected ruptured cyst, which was determined to be parasitic. Due to similarities in appearance, these cysts can be confused with other lesions. Careful management of these cysts in highly prevalent areas must be considered. Despite advanced technology, hydatid cysts present challenges for both surgeons and radiologists.

Keywords: *Echinococcus Granulosus*, Hydatid Cyst, Ovarian Cyst, Ovary

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Introduction

Cases of hydatid cysts cases have been reported worldwide with a diverse prevalence in different regions. The parasite that primarily contributes to its formation in humans is *Echinococcus granulosus*; other species such as *Echinococcus multilocularis* have also been reported (1).

Echinococcus is a type of tapeworm that is usually carried by carnivores such as dogs, which are considered to be the definitive hosts. The eggs laid by these worms are then passed and carried by faeces and they infect herbivores like domestic animals, as the intermediate host. Humans are considered to be incidental intermediate hosts. The duration from cyst formation to clinically symptomatic disease takes around 10 to 20 years. However, this time may change if the primary cyst ruptures (1, 2).

Larvae usually go through metamorphosis in the intestine and then migrate to different parts of the body. Although the usual destination is the liver (63%) and lungs (15%), other unusual locations have been reported (2-4). One of the most peculiar and rare locations of the

cyst is the ovary; it is extremely rare to have a primary cyst in the ovary. Therefore, most cases are the result of secondary cyst formation (2, 4, 5).

Although surgical management is the basic treatment, recurrence rates are as high as 22%. In order to mitigate subsequent recurrences or undesirable consequences, methods such as conservative surgery along with chemotherapy have been proposed (5, 6). It is important to differentiate this cyst from other types of ovarian cysts, particularly malignant ones. Here, we report three cases of ovarian hydatid cyst.

Case 1

A 31-year-old woman with complaints of abdominal colic pain in her right lower quadrant (RLQ) referred to the gynaecology clinic. She came from a rural area near Qom, Iran and had a history of frequent contact with livestock. She had no other complaints or any underlying diseases. The pre-surgery abdominopelvic computed tomography (CT) scan showed a large solid cystic mass in her left ovary that measured 110×86 mm

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with mild enhancement in the solid sections and favoured cystadenoma/adenocarcinoma. The liver and bile ducts were normal. All of the tumour markers were normal, except for CA125, which was mildly elevated at 38.2 U/ml (upper normal limit: 35 U/ml). The patient was scheduled for elective surgery because of the highly suggestive imaging findings and impression of malignancy. However, after one week, she presented to the emergency room with complaints of acute abdominal pain for three days prior. An abdominopelvic sonographic scan revealed a hypoechoic multilocular cyst (88×79 mm) in the left adnexa with an internal hyperechogenic septa and massive free fluid in the Morison pouch, rectouterine pouch, and RLQ. This finding was suggestive for a ruptured ovarian cyst. The patient also had tachycardia. Routine laboratory tests revealed mild anaemia. The patient underwent a laparotomy with a Pfannenstiel incision and a massive amount of ascites and debris were removed. The patient underwent a left salpingo-oophorectomy and the abdominal cavity was subsequently irrigated with saline solution. The patient had an uncomplicated postoperative recovery. The surgical specimen was sent to a pathology laboratory for further evaluation and the result was consistent with a hydatid cyst. Therefore, the patient was referred to an infectious disease specialist who prescribed albendazole. A serologic test for anti-Echinococcus was performed to confirm the diagnosis and she had a positive antibody result.

At the three-month follow-up, another abdominopelvic CT scan showed a hydatid cyst in the left lobe of her liver that had loculated fluid in the left paracolic space and a right adnexal cyst. The cyst was aspirated and puncture of the cyst wall, aspiration of cyst contents, instillation and reaspiration of the scolical agent (PAIR) was performed on the liver cyst. A sonographic scan showed no new pathologic lesions, and only remnants of the cyst were observed in the liver.

Case 2

A 32-year-old woman with a four-year history of infertility and complaints of pelvic pain referred to our clinic because of an ultrasonography report of an adnexal mass. Bimanual pelvic examination showed a palpable pelvic mass. She was previously diagnosed with an ovarian cyst and treated with letrozole and oral contraceptive pill (OCP). However, due to the COVID-19 outbreak, she did not return for her follow-up visits. In the following work up, her transvaginal sonographic scan showed a 10 cm paratubal cyst. All routine laboratory test results were within normal limits. Tumour markers were negative and her Anti-Müllerian hormone (AMH) level was 1.97 ng/ml. The patient underwent laparoscopy for the adnexal mass and it was noted that she had considerable adhesions between the anterior wall and omentum, and a giant cyst. Based on these findings and our experience with the previous patient, we decided to perform a laparoscopic cystectomy. All measures were taken to halt further cyst formation. First, we removed the omental adhesions using

a LigaSure. Then, the intestines were freed posteriorly and the uterus, fallopian tubes, and ovaries were exposed. Gross examination of the excised cyst indicated that it was a hydatid cyst. The germinal layer and cyst wall were removed by hydro-suction, and the sample was retrieved with an endo-bag. The abdominal cavity was irrigated with hypertonic saline and then thoroughly suctioned. A Jackson-Pratt drainage system was subsequently inserted to enable better drainage. After the surgery, the patient had disseminated urticaria; therefore, the anaesthesiologist prescribed chlorpheniramine and hydrocortisone, and the patient was admitted to the ICU where she was prescribed albendazole, the main anti-parasitic medication. Postoperatively, ultrasound (US) evaluation was remarkable for an 80×72 mm cyst in the right lobe of the liver, which was compatible with a hydatid cyst. The patient had an uncomplicated recovery and was discharged on the 14th post-operative day. The patient was also referred to a surgeon for follow-up of the liver cyst.

After surgery, it was determined that she had contact with livestock and dogs, and resided in a rural area near the city of Boroujerd, Iran.

Case 3

A 23-year-old woman (G3P3A1) presented to the clinic with complaints of increasing abdominal pain for two days prior. Her symptoms began during the 10th week of pregnancy. The pain was neither radiating nor migratory, and was localized in the RLQ. Physical examination indicated rebound tenderness. She did not reveal any past medical history at the time of her visit. The patient had a past medical history of previous abortion and prior to her abortion, she underwent surgery to remove a hydatid cyst and was given anti-helminthic therapy (albendazole). After her abortion she believed that the medication was the underlying cause; therefore, she refused to continue albendazole. An abdominopelvic sonographic scan was requested because of her medical history and physical examination findings. The results showed a viable foetus with a foetal heart rate, which was 9 weeks and 5 days old that had crown-rump length (CRL) measurements of 29 cm. Additional findings were the presence of multiple cystic lesions in the right adnexa and ovary, the largest was 50×60 mm and it had an internal isoechoic septum and elements. Another huge extra ovarian cystic lesion (81×21×21 mm) was observed within the right adnexa; this lesion was similar to an ovary in appearance. Massive amounts of free fluid around the right adnexa, rectouterine pouch, and left adnexa were also noted. This report strongly suggested that the findings were compatible with ovarian torsion and there was no detectable blood flow to the cysts. Other tests, such as routine laboratory analyses, revealed only mild anaemia and the tumour markers were within the normal range. Adnexal torsion was the most probable diagnosis; therefore, the patient was scheduled for laparoscopic surgery. Prior to the surgery, the patient informed the staff and the gynaecologist about her past medical history. During the surgery,

attempts were made to investigate the lesions and organs in the abdomen and pelvis. Many loose adhesions were observed within the pelvis and abdomen. The cyst was necrotic and attached to the abdominal wall, but not the ovaries. Her symptoms were attributed to this cyst; therefore, a cystectomy was performed and the general appearance of the cyst was similar to a hydatid cyst. She had numerous cysts disseminated throughout her abdomen and pelvis. The appendix was also examined by the assistant general surgeon and reported to be normal. The abdominal cavity was irrigated with hypertonic saline solution. The obtained specimen was sent to the histopathology laboratory. The patient spent two days in recovery and was later discharged with no complications. The histopathologic report also confirmed hydatidosis. Anti-helminthic therapy was prescribed for the patient; however, she refused the medication as she believed it interfered with her pregnancy. At the time this article was written, two months after surgery, she was pregnant and had no adverse complications (Figs. 1, 2).

Ethical considerations

Our cases were presented to Valiasr Hospital, Imam Khomeini Hospital complex at Tehran University of Medical Sciences. According to the ethical criteria set by Ethical Committee of Tehran University of Medical Sciences in regards to case-reports, we obtained written informed consent forms from all patients.

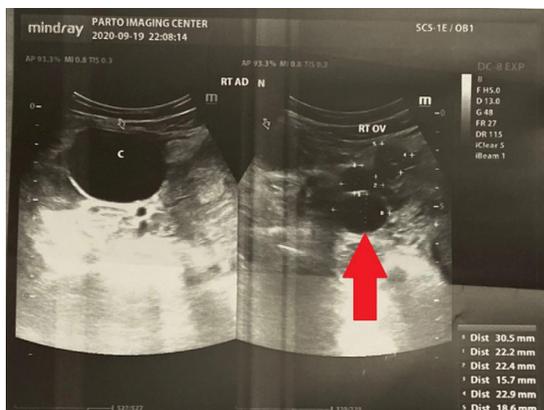


Fig.1: Ultrasonographic image shows the presence of hydatid cysts.

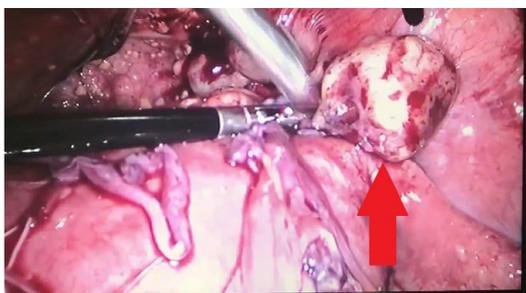


Fig.2: Removal of hydatid cyst during laparoscopic surgery.

Discussion

Hydatid cysts have been reported from many countries and regions of the world; most of the primary cysts are located in the liver or lungs. Therefore, a primary ovarian

hydatid cyst is a rare incidence and most cases with pelvic involvement are secondary to either idiopathic or iatrogenic rupture of the primary cyst (6, 7). Despite numerous technical and technological advancements in both surgical and medical treatment of hydatid cysts, many challenges are encountered to prevent their recurrence. There is no defined consensus for patient follow-up (6).

Due to similar appearances, echinococcal cysts are in differential diagnosis with multilocular cystic adnexal masses and malignancies (8). It is suggested that a diagnosis of hydatid cyst be taken into consideration in highly prevalent areas. The diagnosis is difficult because of non-specific clinical symptoms, lack of sensitive serologic tests, and atypical radiologic findings (4, 8, 9). However, a definitive diagnosis can help with surgery as it reduces the risk for cyst rupture and the spread of protocols (10). The most widely used method for diagnosis is a CT scan or US scan and a previous history of this disease. A vaginal US scan is an important examination that allows for better recognition and diagnosis because of the ability to visualise the cystic or multilocular characteristics, and possibility of visualising the germinal layer (8, 10). A CT scan can help with a better diagnosis because it can show both calcifications and daughter cysts. Although the above-mentioned modalities are highly sensitive, the gold standard for diagnosis is histopathology (4).

However, in the current case series, the patients did not know about their disease status and, because of the rarity of this kind of manifestation, two of our patients were not diagnosed until after surgery and histopathology analysis. Another useful test was the serology test, which was positive in one of the patients.

During the surgery, it is of utmost importance to prevent secondary infection; therefore, all of our patients received hypertonic saline irrigation and, subsequently, anti-helminthic therapy.

Conclusion

Pelvic hydatid cysts, especially ovarian cysts, are rare. Therefore, a high grade of suspicion along with epidemiologic data of the disease and proper use of imaging is required for an accurate diagnosis. This diagnosis must be considered in patients who live in areas endemic for this disease or if they have a previous history of infection. Modalities that contribute the most to diagnosis include US, particularly transvaginal sonography and CT scans. Serologic findings can also be helpful. The optimal treatment is radical cystectomy; however, in some cases, it might not be available. In such circumstances, a partial cystectomy can be useful. In order to prevent subsequent negative outcomes, it is suggested that all patients receive anti-helminthic therapy.

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Authors' Contributions

A.H., A.T.; Participated in study design, data collection and drafting the manuscript. E.I., S.J., R.V., L.H.; Gathered data and took part in draft of manuscript. The data was analyzed and manuscript was revised by A.H., A.T., L.H. All authors participated in final edit of the manuscript.

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