

Primary adnexial hydatid cyst mimicking ovarian tumor

Over tümörünü taklit eden primer adneksiyal kist hidatik

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Abstract

We report here the rare case of a 28-year-old woman with a large hydatid cyst in her left lower pelvis with an unusual sonographic presentation mimicking a multicystic ovarian tumor. Laparoscopic evaluation revealed normal uterus and ovaries with a swelling in the left retroperitoneal area. We decided to reach this tumour by the vaginal route and multiple scolex, daughter cysts were removed via a left lateral vaginal wall incision. The pericystic cavity was thoroughly washed. The patient was discharged on the first postoperative day. Mebendazole (100 mg twice daily) was administered for 4 months. This parasite should be kept in mind and considered when making the differential diagnosis of pelvic cystic masses, particularly if the patient is from an endemic area.

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Key words: Echinococcus, hydatid disease, ovarian tumour

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Özet

Bu vakada 28 yaşındaki genç bir kadında ultrasonografik görüntüsü multikistik over tümörünü taklit eden sol pelvisteki geniş bir kist hidatik vakasını sunuyoruz. Laparoskopik değerlendirilmede uterus ve overler normal iken sol retroperitoneal bölgeye şişlik izlendi. Bu tümöral oluşuma vajinal yoldan ulaşmaya karar verdik ve sol lateral vajinal duvar insizyonu ile birden çok kistik yapıdan oluşan skoleksler ve kitle eksize edildi. Kist çevresi doku tamamen yıkandı. Hasta postoperatif 1. günde taburcu edildi. Dört ay boyunca günde iki kez 100mg mebendazol uygulandı. Özellikle endemik bölgelerden gelen hastalarda kistik pelvik kitlelerin ayırıcı tanısında bu parazit akılda tutulmalıdır. (J Turkish-German Gynecol Assoc 2009; 10: 232-4)

Anahtar kelimeler: Echinococcus, kist hidatik, over tümörü

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Introduction

Hydatid disease (echinococcosis), is a parasitic infection caused by a cestode Echinococcus, mostly the form of *E. granulosus*. Echinococcosis remains a problem in endemic areas. *E. granulosus* is a 5-mm long worm, with a lifespan of 5-20 months within the jejunum of dogs. Echinococcal eggs of the adult worm are present in the small intestine of canine animals, and they are excreted with the feces. When ingested by intermediate hosts such as sheep, cattle or humans, these eggs hatch in the intestine of the intermediate hosts. Then the eggs, in the form of oncospheres, penetrate through the mucosa of the intestine and diffuse into the blood and lymphatic circulation. They are transported by the circulation to the organs, mostly to the liver and lungs, where they grow and produce cysts. Five to 20 years elapse before cysts enlarge sufficiently to cause symptoms.

Echinococcal disease, which produces cystic lesions, is an infection of humans caused by the larval stage of *Echinococcus granulosus*. It is prevalent in the Middle East, the Mediterranean region, particularly in Greece and Lebanon, Australia, Argentina and Africa. Confirmed hosts are dogs that pass eggs into their feces. Intermediate hosts, e.g. sheep, cattle, humans, goats and horses ingest the eggs and develop cysts (1). Echinococcal cysts are mostly found in the liver (60%-70% of cases), followed by the lungs (10%-

25%), spleen, ovaries, kidneys, brain, bones and heart, but rarely elsewhere in the body (1). Hydatid disease in extrahepatic locations usually remains asymptomatic unless the cyst grows and produces symptoms due to pressure, rupture to the pleural or peritoneal cavity, secondary infection, or an allergic reaction (2). We report here the rare case of a 28-year-old woman with a large hydatid cyst in her left lower pelvis with an unusual sonographic presentation mimicking a multicystic ovarian tumour.

Case

A 28-year-old gravida 2, para 2 woman presented at the Department of Obstetrics and Gynaecology with a left adnexial mass on routine examination. The patient described regular menses at 28 day intervals and her last menstrual period was 16 days prior to admission. She had had two term caesarean section deliveries. The gynecological history was otherwise unremarkable. On bimanual examination, a left adnexial cystic mass approximately 10 cm in diameter was palpated. The left-sided lesion was in close proximity to the vagina and left vaginal swelling was also noted. A year previously in a different clinic she had attended with a complaint of lower abdominal pain and was told that she had a cyst. An oral contraceptive containing 30 mg ethinyl estradiol and 3 mg drospironon was administered. She was living in

town and mostly dealing with farm animals for breeding, milking them in close contact. Transvaginal sonography with 7 MHz vaginal transducer revealed a 11x9 cm hypoechogenic multiloculated cystic mass in the left ovarian location. The left ovary could not be visualized separately and the appearance of the cyst mimicked ovarian hyperstimulation syndrome ovaries (Fig.1). On color Doppler no vascularity was noted in the solid component of the mass. She had been asked about any medication for infertility and she used one of them. Tumour markers; CA 125, CA 19-9, CEA, AFP, B-HCG; liver enzymes, biochemical parameters and other hematologic parameters were normal. She was diagnosed as having a benign left adnexial tumour and it was decided to operate by laparoscopy. Laparoscopic evaluation revealed a normal uterus and ovaries with a swelling in the left retroperitoneal area. We decided to reach this tumour by the vaginal route and multiple scolex, daughter cysts were removed via the left lateral vaginal wall incision (Fig. 2-3-4). The pericystic cavity was thoroughly washed with hypertonic saline. The thick pericystic cavity was left open. The diagnosis of a hydatid cyst was confirmed histologically after surgical removal of the lesion. Postoperative CT scan and ultrasound of the liver, spleen and lung were normal, and no disease was

visible on chest radiograph. The patient was discharged on the first postoperative day. Mebendazole (100 mg twice daily) was administered for 4 months.

Discussion

To our knowledge, this is the first reported case of an echinococcus cyst approached by the vaginal route.

The parasite consists of the laminated membrane (ectocyst) and the germinal layer (endocyst). The ectocyst has the appearance of the white of a hard-boiled egg. It is elastic, made up of gelatinous, chitinous material and, when incised or ruptured, curls in on itself, exposing the inner layer. The innermost germinal layer is cellular and consists of a number of nuclei embedded in a protoplasmic mass. It is a very thin, vital layer of the cyst, and produces brood capsules with scolices, secretes

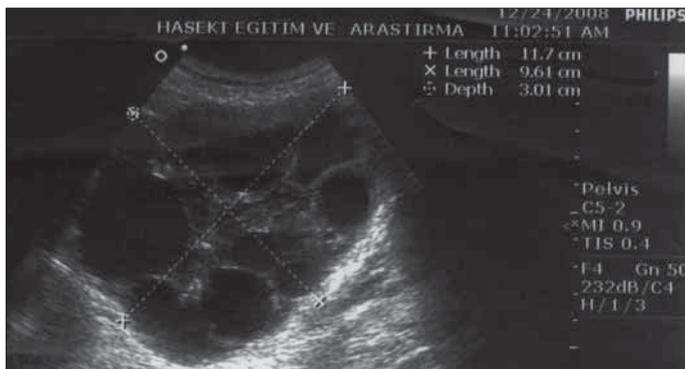


Figure 1. Transvaginal sonography 11x9 cm hypoechogenic multiloculated cystic mass in the left ovarian location



Figure 3. The thick pericystic cavity left open



Figure 2. Tumour by vaginal route and multiple scolex, daughter cysts have been removed via left lateral vaginal wall incision



Figure 4. Multiple scolex, daughter cysts have been removed

hydatid fluid and forms the outer layer. The cyst fluid is crystal clear and colourless with a specific gravity of 1,005 to 1,010, is slightly alkaline, and is highly antigenic and toxic. Contact with the fluid can give rise to anaphylactic shock.

Hydatid cyst affects the liver and the lungs in more than 80–90% of cases. Hydatid cysts are seldom primary in other organs, and they are often part of a generalized disease. Involvement rate of the pelvis in hydatid disease is reported to be 2% (4,5). In females, genital organs are reported to be the most affected areas in the pelvis which can be attributed to their relatively rich bloodstream and true invasions from connective tissue of peritoneum of Douglas and suspensory ligaments (4, 6) Pelvic hydatid disease can present with vague abdominal pains due to irritation, swelling, menstrual irregularities, infertility and pressure symptoms involving the adjacent organs (bladder, ureters, rectum and vascular structures) (3, 4, 7, 8).

Cysts in the peritoneal cavity account for 10%-16% of cases and are mainly the result of the rupture of concomitant hepatic cysts. Extrahepatic locations of the echinococcus include the lungs (10%-15%), spleen (0.9%-8%), kidneys (1%-4%), pancreas (0.25%-0.75%), brain, heart, ovaries, bones and abdominal wall. Symptoms in such cases occur because of pressure or complications including rupture, allergic reaction and secondary infection (9).

Radiography, US and CT studies are important for diagnosis of echinococcal disease. Plain abdominal X-rays may show calcifications of the cystic wall (10). US is the method of choice for the detection of hepatic and extrahepatic echinococcal cysts.

Hydatid cysts are classified by ultrasound into six categories. Type 1 are defined as univesicular and are <50 mm in diameter. Type 2 are univesicular with a prominent laminated layer, and tend to be seropositive. Type 3 are subdivided into 3a, defined as cysts with a prominent lamination that contains daughter cysts, and 3b, characterized by lamination but a lower number of daughter cysts. Both 3a and b are highly seropositive. Type 4 appear as solid masses. Type 5 are characterized by degeneration with calcifications. Type 6 are defined as multiple cysts that may be univesicular and laminated, with daughter cysts involving one or more organs (11, 12). The sensitivity of US ranges from 93% to 98% (12).

Serological tests contribute to diagnosis. Immunoglobulin G antibody detection by ELISA has a sensitivity of 95% and a specificity of 94%. The sensitivity of indirect hemagglutination test has been found to be 87.5% (11).

Primary pelvic hydatid cyst, although rare, can be found in association with liver or lung hydatid cysts. Complications of pelvic hydatid disease may be urinary problems, rupture, or even obstructed labor (5, 7, 13-15).

The World Health Organisation has outlined the treatment guidelines for hydatid cysts. Surgery is the treatment of choice for all patients with symptomatic disease and who are fit for surgery (16). For symptomatic or large hydatid peritoneal cysts, surgery, when feasible, is the principal method of treatment. Surgical treatment can be either radical or conservative. Total cystectomy, whenever possible, is the gold standard. For peritoneal cysts firmly attached to intraperitoneal viscera, unroofing and drainage has been proven to be a safe method. It is important that the abdominal cavity is isolated with gauzes soaked in 20% hypertonic saline solution to avoid secondary hydatidosis and allergic reaction (17).

Generalized toxic reaction due to hydatid cyst rupture and secondary infection are among the most common complications. The evacuation of the pelvic cyst by the vaginal route may prevent the risk of toxic reaction.

The pelvic hydatid cyst in the case of Gupta et al. (6) was located in the right pelvis, and it extended through the greater sciatic notch into the gluteal region. The recurrence incidence of hydatid disease after surgery is said to be 8-22%, with recurrences most often noted within 2 years after the operation (3). Physicians should be familiar with hydatid cyst imaging features and direct the surgeon, so that care can be taken during operation in order to prevent recurrences and choose the appropriate treatment options. This parasite should be kept in mind and considered when making the differential diagnosis of pelvic cystic masses, particularly if the patient is from an endemic area. The vaginal approach and evacuation of the cyst seems safe, effective and prevents the toxic reaction.

References

1. Tsaroucha AK, Polychronidis AC, Lyrantzopoulos N, Pitiakoudis MS, J Karayiannakis A, Manolas KJ, Simopoulos CE. Hydatid disease of the abdomen and other locations. *World J Surg* 2005; 29: 1161-5
2. Prousalidis J, Tzardinoglou K, Sgouradis L, Katsohis C, Aletras H. Uncommon sites of hydatid disease. *World J Surg* 1998; 22: 17-22
3. Pedrosa I, Saiz A, Arrazola J, Ferreiros J, Pedrosa CS. Hydatid disease: radiologic and pathologic features and complications. *Radiographics* 2000; 20: 795-817
4. Terek MC, Ayan C, Ulukus M, Zekioglu O, Ozkinay E, Erhan Y. Primary pelvic hydatid cyst. *Arch Gynecol Obstet* 2000; 264: 93-6
5. Hassan FO, Shannak A. Primary pelvic hydatid cyst: an unusual cause of sciatica and foot drop. *Spine* 2001; 26: 230-2
6. Gupta A, Kakkar A, Chadha M, Sathaye CB. A primary intrapelvic hydatid cyst presenting with foot drop and a gluteal swelling. *J Bone Joint Surg* 1998; 80: 1037-9
7. Seenu V, Misra MC, Tiwari SC, Jain R, Chandrashekhar C. Primary pelvic hydatid cyst presenting with obstructive uropathy and renal failure. *Postgrad Med J* 1994; 70: 930-2
8. Solidoro G, Del Gaudio GA. Hydatid cysts of the ovary. Description of a case. *Minerva Chir* 1991; 46: 571-5
9. Hamamci EO, Besim H, Korkmaz A. Unusual locations of hydatid disease and surgical approach. *ANZ J Surg* 2004; 74: 356-60
10. Sayek I, Onat D. Diagnosis and treatment of uncomplicated hydatid cyst of the liver. *World J Surg* 2001; 25: 21-7
11. Balik AA, Celebi F, Basglu M, Oren D, Yildirgan I, Atamanalp SS. Intra-abdominal extrahepatic echinococcosis. *Surg Today* 2001; 31: 881-4
12. Shambesh MA, Craig PS, Macpherson CN, Rogan MT, Gusbi AM, Ehtuish EF. An extensive ultrasound and serologic study to investigate the prevalence of human cystic echinococcosis in northern Libya. *Am J Trop Med Hyg* 1999; 60: 462-8
13. Ladeb MF, Bouhaoula H, Slim K, Ganouni A. Pelvic hydatidosis in women. Apropos of 3 cases. *J Gynecol Obstet Biol Reprod* 1989; 18: 493-5
14. Arousseau R, Martinon F. Single ruptured pelvic hydatid cyst as an unusual manifestation of secondary peritoneal echinococcosis. *J Chir* 1977; 114: 167-74
15. Solidoro G, Del Gaudio GA. Hydatid cysts of the ovary. Description of a case. *Minerva Chir* 1991; 46: 571-5
16. Guidelines for treatment of cystic and alveolar echinococcosis in humans. WHO Informal Working Group on Echinococcosis. *Bull World Health Organ* 1996; 74: 231-42.
17. Karavias DD, Vagianos CE, Kakkos SK, Panagopoulos CM, Androulakis JA. Peritoneal echinococcosis. *World J Surg* 1996; 20: 337-40