CASE REPORT

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Primary Uterine Hydatid Cyst Incidentally During Cesarean Section

Hale ÇETİN ARSLAN[a]

[a]University of Health Sciences Faculty of Medicine, Kanuni Sultan Süleyman Training and Research Hospital, Clinic of Obstetrics and Gynecology, Department of Perinatology, İstanbul, Türkiye

ABSTRACT Cystic echinococcosis is a zoonosis known as hydatid disease transmitted from animals to humans. It is most commonly seen in the liver and lungs, and genital organ hydatid cyst cases are rarely reported in the literature. The incidence of hydatid cysts detected during pregnancy is 1 in 20,000-30,000. Although it is so rare, diagnosis and effective treatment are highly critical to the health of the mother and the newborn. In this case, which is considered to be a primary uterine hydatid cyst detected in a term pregnant woman who applied to our clinic for delivery, we aim to share our experience with the unusual localization of hydatid cysts and our successful management and significantly to help obstetricians keep this in mind in the differential diagnosis.

Keywords: Adnexa uteri; echinococcosis; pregnancy

Hydatid cyst is an endemic zoonotic disease caused by the *Echinococcus* parasite.[1] The most common species infecting humans is *Echinococcus granulosus* and *Echinococcus multilocularis*. This chronic disease is endemic, especially in agriculture and animal husbandry societies. In Türkiye, the prevalence is assumed to be 50 per 100,000, and the incidence is supposed to be 2 per 100,000.[2] The fecal-oral route transmits it. It can be seen almost anywhere in the human body, most commonly in the liver (75%) and lungs (15%). Pelvic localization usually develops secondary to the rupture of a liver cyst, and primary female genital organ involvement is quite rare. In our patient, who was diagnosed randomly during cesarean section and had no additional complaints other than labor pain, no additional location of the hydatid cyst outside the uterus was detected in the imaging. During pregnancy, hydatid cysts can grow and spread rapidly due to the suppression of cellular immunity and steroids secreted from the placenta. Rupture of the cyst content can cause complications such as deterioration of the general condition, anaphylactic shock, and bleeding, which can be life-threatening for both the mother and the baby. The rare localization of hydatid cyst cases, which general surgeons more frequently see, makes diagnosis somewhat tricky for clinicians. This case, which is the first seen in our clinic, has no specific history or chronic disease in her history, and the hydatid cyst diagnosis was made incidentally during a cesarean section, which is quite unusual for us.

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Correspondence: Hale ÇETİN ARSLAN
University of Health Sciences Faculty of Medicine, Kanuni Sultan Süleyman Training and Research Hospital, Clinic of Obstetrics and Gynecology, Department of Perinatology, İstanbul, Türkiye
E-mail: halecetin90@gmail.com

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A 23-year-old gravida 6, parity 5, term pregnant woman with a history of spontaneous vaginal delivery presented to the emergency room with labor pains. The patient stated in her medical history that she worked with scrap metal and had no known chronic disease. Abdominal ultrasonography revealed a cephalic presentation and a live intrauterine singleton pregnancy compatible with the term. Vaginal ultrasonography revealed a 5x5 cm diameter polycystic mass lesion extending from the posterior uterus to the cervix, multiloculated and containing septations (Figure 1). A vaginal examination revealed 4 cm cervical dilatation and a palpable immobile mass. The patient was admitted to the delivery room for delivery. A general surgeon's opinion was requested regarding the nature of the mass. Based on the medical history and ultrasonographic images, a hydatid cyst was considered in the preliminary diagnosis. Magnetic resonance imaging (MRI) was requested. However, it could not be performed due to the patient's increasing pain and the progress of labor. A decision was made to consult the general surgery and perform a cesarean section to prevent possible rupture of the cystic mass and to avoid an anaphylactoid reaction. When the abdomen was entered, 5-6 well-defined, complete cystic masses were seen in the posterior part of the uterus, including the omentum (Figure 2). The cysts were easily excised from the surrounding tissues. There were many cysts with a thick consistency, gelatinous material in places, and irregular roughness on the inner surfaces of the removed masses. It was considered a stage 2-3 hydatid cyst (Table 1).[3] Cuticle layer and protoscolices were observed in the cysts sent for histopathological examination (Figure 3). Upon the recommendation of the infectious diseases department, an anthelmintic treatment protocol (Albendazole 400 mg 2*1) was started, and postoperative brain, abdomen, and thorax tomography imaging was requested; no abnormality or additional involvement area was detected. The indirect hemagglutination test result was reported as 1/2,560. The case was interpreted as a primary uterine hydatid cyst. The patient, who did not develop any complications during the follow-up period, was discharged to come for infectious disease control. Written consent was ob-

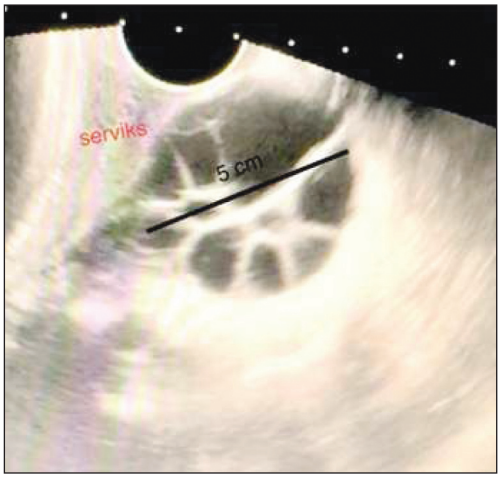

FIGURE 1: Cyst appearance on vaginal ultrasonography

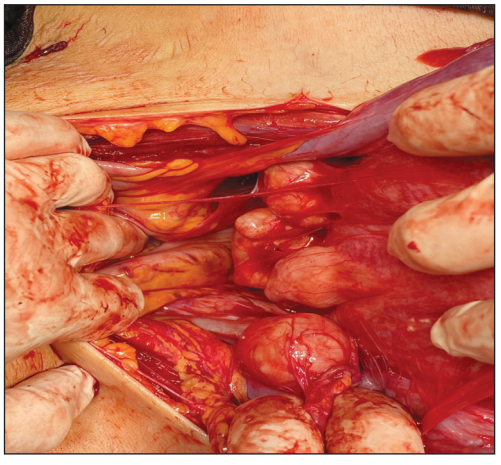

FIGURE 2: Extensive cyst localization in the abdominal and uterine posterior wall during cesarean section

TABLE 1: Gharbi classification of cystic echinococcosis

CE 1-active stage	• Unilocular anechoic cyst with double line sign • Delicate internal echoes represent "hydatid sand" (fluid and protoscolices originating from a ruptured vesicle)
CE 2-active stage	• Cyst with multiple internal septations • Septa represent walls of daughter cyst(s) • Described as multivesicular, rosette, or honeycomb appearance
CE 3-(transitional stage: the evolving appearance of daughter cyst(s) within the encompassing parent cyst	• 3A: Daughter cysts have detached laminated membranes (water lily sign) • 3B: Daughter cysts within a solid matrix
CE 4-(inactive/degenerative)	• Cyst with heterogeneous hypoechoic and hyperechoic contents (ball of wool sign) • Absence of daughter cysts
CE 5-(inactive/degenerative)	• Solid and calcified wall

CE: Cystic echinococcosis

FIGURE 3: Completely excised hydatid

tained from the patient to publish the patient's clinical details and images, ensuring that the patient's identity would not be disclosed.

DISCUSSION

The *Echinococcus* parasite causes encapsulated, non-invasive cystic lesions in the affected organ. It has four forms: *Echinococcus granulosis, E. multilocularis, E. vogeli* and *E. oligarthrus* (rare in humans). Rare localizations of hydatid cysts such as brain, heart, pericardium, kidney, bone, and breast tissue have always been curious.[4] Hematogenous or intestinal lymphatics mainly spread it. It is also encountered primarily by cyst rupture or the parasite spreading with intraperitoneal fluid and seeding other organs. Usually, a single organ is affected; 2 organ involvement is detected in 10-15% of cases.[5] Primary uterine hydatid cyst cases are still being discussed in the literature. Pelvic involvement is between 0.2-0.9%, and the ovary is most frequently affected, followed by the uterus. In cases of ovarian involvement, patients usually present with complaints such as pain and abdominal distension, and symptoms vary depending on the location of involvement. Our case, however, does not express a symptom specific to the mass since she was pregnant, which made it difficult for us to make a diagnosis.

The first primary uterine hydatid cyst case was published in 1847.[6] In later years, there are also case reports of a patient who underwent surgery due to the presence of a mass in the uterus and was initially thought to be malignant, and whose final pathology result was reported as a hydatid cyst.[7] It should always be remembered that the disease can affect multiple organs. A patient diagnosed with a hydatid cyst should undergo a systemic examination, primarily of the liver and lungs, and serological tests and imaging techniques (ultrasound, tomography, and MRI) should be included in the examination, especially to detect atypical localizations. The gold standard diagnostic method is histopathological examination. Hydatid cysts are generally in the form of a single-loculated cyst and rarely in the form of a multiloculated cyst. A 3-layered cyst wall is seen in microscopic examination. The pericyst layer neutralizes the host's response to the parasite; the white and cell-free laminated membrane forms the intermediate layer, and the transparent and thin inner layer forms the germinal layer.[8]

The incidence of hydatid cysts during pregnancy is 1/20,000. Even if pregnant women are asymptomatic, they may become symptomatic due to decreased cell-mediated immunity.[9] Although there is no standard surgical or medical treatment, the gold standard treatment is complete surgical excision of the cyst.[10] To prevent postoperative recurrence and high-risk contamination and in the presence of multiple localizations, benzimidazole derivatives (albendazole, mebendazole) are frequently used.[11] However, it should be known that the pregnancy category of these treatments is C.

The rarity of uterine hydatidosis, its indolent course, the heterogeneity of symptoms, and the clinician's failure to consider it in the differential diagnosis make preoperative diagnosis difficult. In pelvic masses, cystic or mixed retroperitoneal tumors (dermoid cysts), pyogenic or tuberculous abscesses, ovarian or hydrosalpinx cysts, ovarian tumors or uterine myomas, and hydatid cysts should be kept in mind in the differential diagnosis.[12] Hydatidosis continues to be a public health problem, especially in endemic and underdeveloped communities, and preventive measures are required primarily to prevent human contamination.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

This study is entirely author's own work and no other author contribution.

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