

Primary Uterine Hydatid Cyst: Case Report

Primer Uterin Kist Hidatik: Olgu Sunumu

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ABSTRACT

Hydatid cyst is a parasitic condition that can be detected practically everywhere in the body. They can cause a variety of symptoms and indications depending on where they are involved in the body. Cestodes termed echinococcus granulosus, echinococcus multilocularis, echinococcus oligarthrus, and, more infrequently, echinococcus vogeli cause the sickness.

This condition, which is widely recognized among general surgery professionals in our country, continues to pose a challenge due to our nation's evolving and changing socioeconomic structure, as well as the fact that it can occasionally be clinically and radiologically mistaken with malignancies.

A 55-year-old female patient with stomach discomfort was admitted to the obstetrics and gynecology outpatient clinic. Her personal history revealed no evidence of a hydatid cyst. Imaging procedures revealed a mass in the patient's uterus. Total abdominal hysterectomy and bilateral salpingo-oophorectomy were performed at the obstetrics and gynecology service with a pre-diagnosis of malignancy. The diagnosis of hydatid cyst was obtained via a microscopical histomorphological investigation. We will report a rare instance of primary uterine hydatid cyst in our investigation.

Key Words: Uterine hydatid cyst, E. granulosus, echinococcosis, primary uterine, hydatidosis.

ÖZET

Kist hidatik, vücudun neredeyse her yerinde görülebilen parazitik bir hastalıktır. Vücuttaki tutulum yerlerine göre farklı semptomlar ve bulgular verebilirler. Hastalığın etkenleri ise echinococcus granulosus, echinococcus multilocularis, echinococcus oligarthrus, daha nadir olarak da echinococcus vogeli adlarındaki sestodlardır.

Ülkemizde genel cerrahi uzmanlarının çok iyi bildiği bu hastalık, ülkemizin gelişen ve değişen sosyal yapısından ve zaman zaman klinik ve radyolojik olarak malignitelerle karışabileceğinden hala sorun teşkil etmektedir.

55 yaşındaki bir kadın hasta karın ağrısı şikayetiyle, kadın doğum ve hastalıkları polikliniğine başvurdu. Özgeçmişinde kist hidatik öyküsü bulunmayan hastanın uterusunda, görüntüleme yöntemleri ile kitle saptandı. Kadın doğum ve hastalıkları servisinde malignite ön tanısıyla histerektomi ve bilateral salpingooferektomi uygulandı. Mikroskopik olarak yapılan histopatolojik incelemede ise kist hidatik tanısı konuldu. Çalışmamızda nadir görülen primer uterin kist hidatik vakasını sunacağız.

Anahtar Sözcükler: Uterin hidatik kist, E. granulosus, ekinokokkozis, primer uterus, hidatik kist, hidatidoz.

INTRODUCTION

The zoonotic illness hydatid cyst is caused by the larval form of Echinococcus. Echinococcosis granulosus larvae infect humans as an intermediate host, causing cystic lesions that are most commonly detected in the liver (50-54 percent) and the lungs (35-40 percent) (1). Other organ involvement may be secondary to involvement of the liver or lungs, or it may be primary (2).

Cestodes that generate hydatid cysts are most widespread in rural Peruvian Andean locations, with a human prevalence of 2-6 percent (3). In the same area, hydatid cyst agent was found in 46 percent of dogs (3). Mongolia and Kazakhstan have the highest frequency of hydatid illness in Asia. The incidence of hydatid illness in our nation is reported to be 0.0034 percent (1). Although it may be found in all locations, it is most common in Eastern Anatolia, Central Anatolia, Marmara, and Thrace. It is noticed 2-6 times more commonly among those living in rural areas than in

those living in urban areas, rather than a difference across regions (4). Although E. granulosus is the most frequent in our nation, instances of E. multilocularis have also been observed.

We present a case of a primary uterine hydatid cyst identified with pelvic cancer using imaging techniques.

CASE REPORT

A 55-year-old lady presented to the obstetrics and gynecology outpatient clinic with a 2-month history of stomach discomfort. Ultrasonography indicated a cystic lesion at the uterine corpus, intramural, on the posterior wall, with exophytic expansion towards the lumen, approximately 7 cm in diameter, with thick walls and septations. The uterus was big according to the age of the patient on magnetic resonance imaging, measuring 74x48 mm in its widest region, hyperintense on T2-weighted sequences, hypointense on T1-weighted sequences, and multiseptal cystic development was noted (Figure 1).

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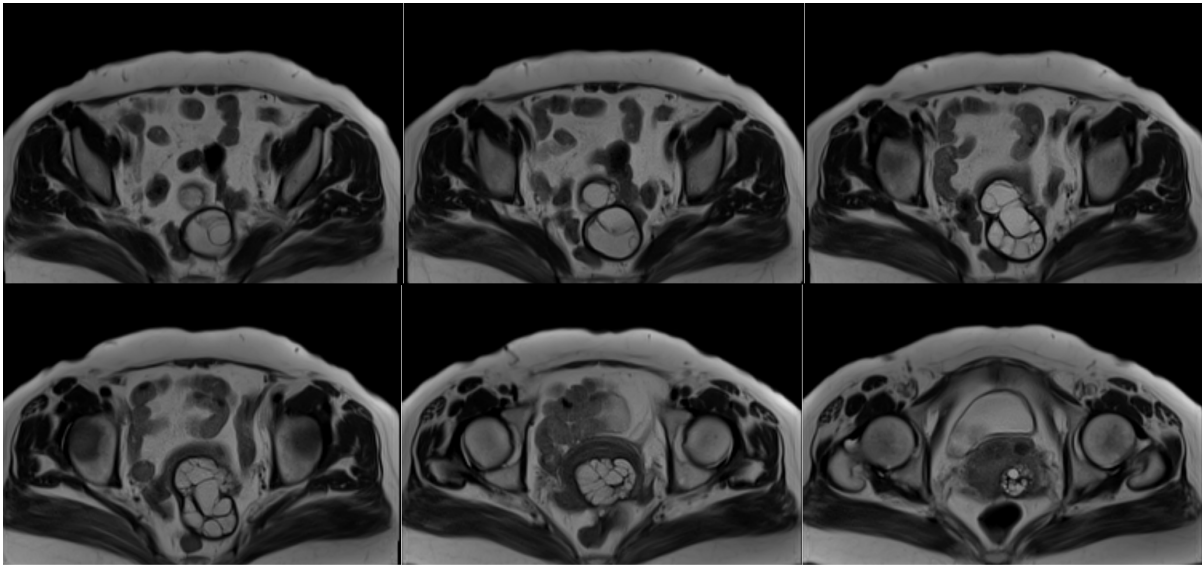


Figure 1. T2-weighted MR imaging reveals hyperintense multiseptal cystic development.

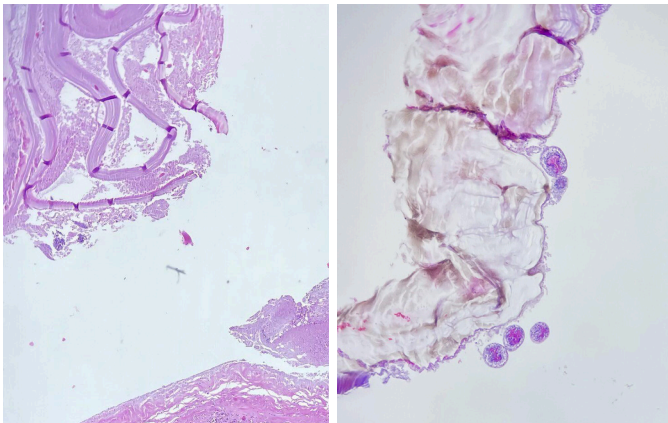


Figure 2. Free-standing cuticular membrane, cyst wall structure that totally atrophies the endometrium, invades the myometrium, and induces an inflammatory response (x40 HE).

Figure 3. Germinative membrane with scolex structures (x100 HE).

On imaging, the other organs were normal. With the suspicion of malignancy, the patient was admitted to the gynecology and obstetrics service. Tumor markers and other blood parameters were found to be within normal limits in blood testing. The patient with a preliminary diagnosis of malignancy underwent total abdominal hysterectomy and bilateral salpingo-oophorectomy (TAH-BSO). There were no complications following the procedure.

A 6x4x4 cm white multilocular cyst formation centered in the fundus and extending to the cervix was detected in the uterine section during macroscopic inspection of the TAH-BSO material sent for pathological assessment. Pearlescent, degradable membrane structures were found on the cyst's cross-sectional surface. A cystic lesion filled the uterine cavity, fully compressing the endometrium and causing myometrium thinning, according to microscopic inspection (Figure 2). Hematoxylin and eosin staining revealed scolex-containing cuticular and germinative

membranes (Figure 3). There is granulomatous inflammation of the foreign body type in the myometrium in response to the lesion. The ovaries and tuba uterinae were both normal. The case was identified as uterine hydatid cyst based on these histomorphological characteristics, which are pathognomonic for the diagnosis of hydatid cyst. Following the diagnosis, the patient was examined for hydatid cyst. During the physical examination, no masses were seen in the extremities. Previous studies revealed that the liver and lungs were clean, thus no extra investigation was required. The hydatid cyst looks intact on magnetic resonance imaging of the abdomen, pelvis, and thorax.

2 months after the procedure, a hydatid cyst serology (1/1280) was found in the blood. In addition, computed tomography was used to image the brain. The exams revealed no evidence of hydatid cysts. The patient was given albendazole for 28 days. She experienced no return of hydatid illness during a one-year follow-up.

DISCUSSION

Hydatid cyst, commonly known as hydatidosis or echinococcosis, is a cyclo-zoonotic infection caused by the cestodes (bandworms) *Echinococcus granulosus*, *Echinococcus multilocularis*, *Echinococcus vogeli* and *Echinococcus oligarthrus*. More common than the others are *Echinococcus granulosus* and *Echinococcus multilocularis*. The unilocular cystic type of hydatid cyst generated by *E. granulosus* is far more prevalent than the uncommon multilocular variant caused by *E. multilocularis* (2).

Dogs, wolves, and foxes serve as definitive hosts, whereas sheep, cattle, and horses serve as intermediate hosts. Since we are only intermediary hosts, we have no function in the biological cycle. Hydatid cyst is thus endemic in

areas of the world where cattle and sheep are bred. Oral consumption of food or water tainted with echinococcus eggs causes human infection (2).

Hydatid cysts can be seen in the pelvic organs in addition to the liver, which accounts for 50–54 percent of their involvement. Pelvic organ involvement ranges from 0.3 percent to 4.27 percent and is uncommon (5). The ovaries and uterus are the most commonly involved pelvic organs (6). For the first time, a uterine hydatid cyst was diagnosed in our clinic.

In 20-30% of instances, several organs are involved. As a result, individuals with hydatid cysts in any area of the body should have a full systemic evaluation, including the liver and lungs (2). No hydatid cyst was found in any organ other than the uterus during the imaging and examination of our patient.

In 1847, Gordon published the first example of a primary uterine hydatid cyst in the literature (7). Giles in 1911 and Hagberg and Maizels in 1954 both met primary uterine hydatid cysts in the literature (8, 9). Kakaie et al. described an Iranian instance in which the primary involvement was the uterine (10). Ennaceur et al. reported two cases of hydatid cyst in Tunisia, the primary site of involvement was the uterus and one was prediagnosed as serous cystadenoma (11). According to the case report by Loukil and Zouari, hydatid cysts were found in the liver and uterus (12). Okumuş, on the other hand, documented a case in which the uterus was the primary involvement (13). Başgül et al. described a case of uterine hydatid cyst formation following hepatic hydatid cyst formation (14). Nermin Koç described a case of a primary uterine hydatid cyst that mimicked a uterine leiomyoma (15). Peker et al. described a case of hydatid cyst with the primary site being the uterine (16). The uterus was the primary site of involvement in our case.

Because of the resemblance between hydatid cyst and malignant illness of the associated organ, accurate diagnosis is critical. As a result, hydatid cysts should be included in the differential diagnosis of cystic pelvic masses, particularly in endemic areas (14, 15). Imaging modalities revealed a multiseptal cystic development originating from the uterine fundus and spreading to the cervix in our case. The patient was operated on with a preliminary malignancy diagnosis.

The hydatid cyst is normally solitary and unilocular on macroscopic inspection. However, like in our case, multiloculated cyst form is uncommon. Histopathological investigation is the gold standard for determining the

presence of a hydatid cyst. The three layers of the cyst wall are visible under a microscope. The pericyst layer is a strong protective layer that limits the host's reaction to the parasite. A white acellular laminated membrane forms the intermediate layer. The inner germinal layer is transparent and thin (17). In our situation, these three layers were also detected. There are around 20-25 case reports of primary hydatid cyst in the uterus in the entire literature study (7-11, 13-16, 18-22). Our case, both macroscopically and microscopically, is consistent with a hydatid cyst and is comparable to others described in the literature.

The gold standard therapy is complete surgical excision of the hydatid cyst (23). Postoperative benzimidazole derivatives (albendazole, mebendazole) are frequently used to avoid recurrence and high-risk contamination (24).

CONCLUSION

We'd like to point you that hydatid cysts are occasionally mistaken for pelvic or genital cancers. Despite sensitive imaging tools, clinical diagnosis of hydatid cyst is not always possible, and it might be mistaken with malignancies. As a result, in the differential diagnosis of pelvic masses, physicians and radiologists should include hydatid cyst.

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REFERENCES

1. Durgun C, Alkan S, Durgun M, Demiray Dindar EK. Türkiye'den Kist Hidatik Konusunda Yapılmış Yayınların Analizi. Black Sea Journal of Health Science. 2022;5:45-9.
2. Goyal P, Ghosh S, Sehgal S, et al. Primary multilocular hydatid cyst of neck with unique presentation: a rare case report and literature review. Head Neck Pathol. 2014;8:334-8.
3. Gavidia CM, Gonzalez AE, Zhang W, et al. Diagnosis of cystic echinococcosis, central Peruvian Highlands. Emerg Infect Dis. 2008;14:260-6.
4. Çobanoğlu U. Tarihçe ve Epidemiyoloji. In: Yalçinkaya İ, editor. Akciğer Hidatik Kisti. Van: Türkiye Solunum Araştırmaları Derneği; 2016. p. 15.
5. Ben Ismail I, Zenaidi H, Rebi S, Zoghalmi A. Primary hydatid cyst of the fallopian tube. IDCases. 2020;20:e00790.
6. Jafarian A, Fakhar N, Parsaei R. Hydatid cyst of fallopian tube. Ann Med Health Sci Res. 2014;4:S324-5.
7. Gordon FH. A case of uterine hydatids. The Western Journal of Medicine and Surgery 1847;7:9.
8. Hagberg CJ, Maizels G. Solitary hydatid cyst of the uterus. S Afr Med J. 1954;28:499-500.

9. Giles A. A case of hydatid of the uterus. *The Lancet*. 1911;177:1700.
10. Kakaei F, Asvadi Kermani T, Tarvirdizade K. A case report: Primary hydatid cyst of uterus. *Int J Surg Case Rep*. 2018;42:67-9.
11. Ennaceur F, Toumi D, Jaouad F, et al. Primary Echinococcus Hydatid Cyst of the Uterus: An Unusual Location. *Case Rep Surg*. 2021;2021:9977326.
12. Loukil I, Zouari A. [Hydatid cyst of the uterus: a rare localization]. *Pan Afr Med J*. 2021;39:272.
13. Okumus Y, Tayyar M, Patiroglu T, Aygen E. Uterine hydatid cyst. *Int J Gynaecol Obstet*. 1994;45:51-3.
14. Basgul A, Kavak ZN, Gokaslan H, Kullu S. Hydatid cyst of the uterus. *Infect Dis Obstet Gynecol*. 2002;10:67-70.
15. Koc N. Primary Hydatid Cyst Mimicking Uterine Leiomyoma. *Turkiye Parazitol Derg*. 2017;41:57-9.
16. Peker K, Ulug P, Nayki UA, et al. Primary uterine hydatid cyst: a case report. *Turkiye Parazitol Derg*. 2013;37:302-4.
17. Sultana N, Hashim TK, Jan SY, Khan Z, Malik T, Shah W. Primary cervical hydatid cyst: a rare occurrence. *Diagn Pathol*. 2012;7:157.
18. Langley G. Primary echinococcal cyst of the uterus. *British Journal of Surgery*. 1943;30:278-80.
19. Jamwal S, Manhas A, Manhas K. Hydatid cyst of uterus-a case report. *Indian Journal of Basic and Applied Medical Research*. 2016;5:104-5.
20. Malik AH, Bashir S, Rather AA. Ruptured primary uterine serosal hydatid cyst: a rare case report. *Gynecological Surgery*. 2005;2:25-6.
21. Dhaifalah I. Hydatid cyst of the uterine cervix. *Biomed Pap Med Fac Univ Palacky Olomouc Czech Repub*. 2001;145:77-8.
22. Kilinc N, Arican C. A hydatid cyst found in an uncommon site coincidentally. *Pakistan Journal of Medical Sciences*. 2007;23:774.
23. Dziri C, Haouet K, Fingerhut A, Zaouche A. Management of cystic echinococcosis complications and dissemination: where is the evidence? *World journal of surgery*. 2009;33:1266-73.
24. Tekin M, Osma U, Yaldiz M, Topcu I. Preauricular hydatid cyst: an unusual location for echinococcosis. *Eur Arch Otorhinolaryngol*. 2004;261:87-9.

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