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Case Report

Multiorgan Echinococcosis with Uterine Involvement Causing Bilateral Hydronephrosis in a Child: Case Report

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Received 12 Mar 2022
Accepted 10 May 2022

Keywords:
Hydatid cyst;
Uterus;
Multiorgan;
Children;
Hydronephrosis,
Turkey

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Abstract

Hydatid cyst is a parasitic infection transmitted by oral ingestion of *Echinococcus granulosus* eggs. Hydatid cyst of the genital tract is rare and the occurrence in the uterus is an extreme rarity. We present an 8-yr-old girl with complaints of swelling of lower abdomen, pollakiuria and bilateral flank pain was brought to Emergency Department of Harran University, Turkey, in Jun 2019. The patient had simultaneous hydatid cysts of the liver, mesentery and uterus. We performed abdominal exploration and completely removed the inner germinal layer of cyst through an incision made in the anterior of the uterine fundus. Then, we applied total excision to the two cysts in the right and left colon mesentery. Finally, we performed partial cystectomy to the cyst in the liver, and we removed the cyst membrane totally. In endemic regions, hydatid cysts should be considered for the diagnosis of children with cystic mass lesions. Uterine-sparing approach should be kept in mind as an option, especially in young women. Early surgical treatment of large pelvic cysts that cause obstructive uropathy may prevent the progression of renal damage.

Introduction

Hydatidosis, also known as cystic echinococcosis (CE), is a zoonotic disease, caused by an infection with *Echinococcus granulosus* parasite especially in endemic areas (1). Although liver and lung are the most common sites, 10% involvement may occur in other parts of the body. Female pelvic organ involvement is ex-

tremely rare and ranges from 0.2 to 4.27% in the literature (2). These atypical sites are generally secondary to spontaneous or traumatic rupture of the cyst in the liver or surgical implantation (1).

There are a few cases of hydatid cyst in adult age group reporting gynecological organ involvement such as ovary, fallopian tube and uterus in the literature (2). The ovaries are the most commonly involved pelvic organ in women and the uterus is the second (3). There is only one reported case of an ovarian hydatid cyst in childhood (4). To our knowledge, our case is the first uterine hydatid case under 16 yr of age ever reported.

We present an 8-yr-old case with liver, mesentery and uterine cyst hydatid simultaneously. Not only being the first uterine hydatid case in the prepubertal period, our case had an interesting course for being symptomatic due to the compression on the urinary tractus.

Case Report

An 8-yr-old girl with complaints of swelling of lower abdomen, pollakiuria and bilateral flank pain was brought to Emergency Department of Harran University, Turkey, in Jun 2019. On abdominal examination, a suprapubic swelling was observed (Fig. 1. A).

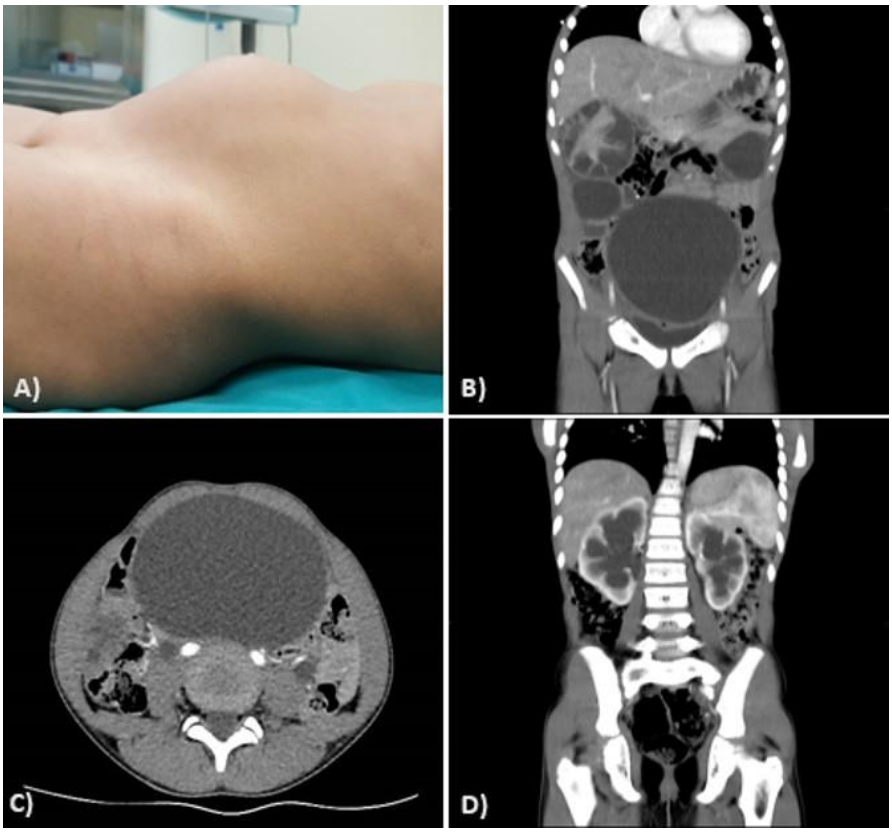

Fig. 1: Images of abdominal swelling and Computed Tomography (CT). A) The appearance of abdominal swelling, B) Image of all cysts in coronal section of CT, C) Pelvic cyst in transverse section of CT, D) Bilateral hydronephrosis in coronal section of CT

There was no history of pelvic or abdominal surgery. Abdominal ultrasonography revealed a 13 x 9 cm unilocular cystic lesion extending into the pelvic region. In addition, 70x58 mm septated and multilobulated cyst appearance compatible with Gharbi type 3 cyst hydatid

was reported in liver segment five localization. There was no free fluid in the abdomen.

Computed tomography revealed a 64x56 mm honeycomb-like septal and lobulated contoured hypodense lesion in segment 5 of liver, two mesenteric cysts (measuring 46x44 mm in the right and 40x37 mm in the left) (Fig. 1.B). A hypodense cyst with a size of 130 x 111 x 72 mm, extending from posterior of bladder to the superior level of umblicus (Fig. 1.C). These cysts in mesenter and pelvic region were all Gharbi stage 1-hydatid cysts. Both bladder and distal level of ureters were compressed due to the pelvic cystic mass. Bilateral Grade 3 hydronephrosis was present (Fig. 1.D). The patient lived on a rural farm with a typical animal contact history (dog and sheep). The preoperative diagnosis was synchronous liver, mesenteric and pelvic HD.

The remainder of the physical and laboratory analysis, such as blood pressure, pulse, fasting plasma glucose, electrolytes, liver and renal function tests, urine analysis and chest X-ray were all normal.

We performed laparotomy the day after patient's admission. Abdominal exploration was performed through a midline incision. We unexpectedly noticed that the largest cyst originated from the uterus. (Fig. 2. A).

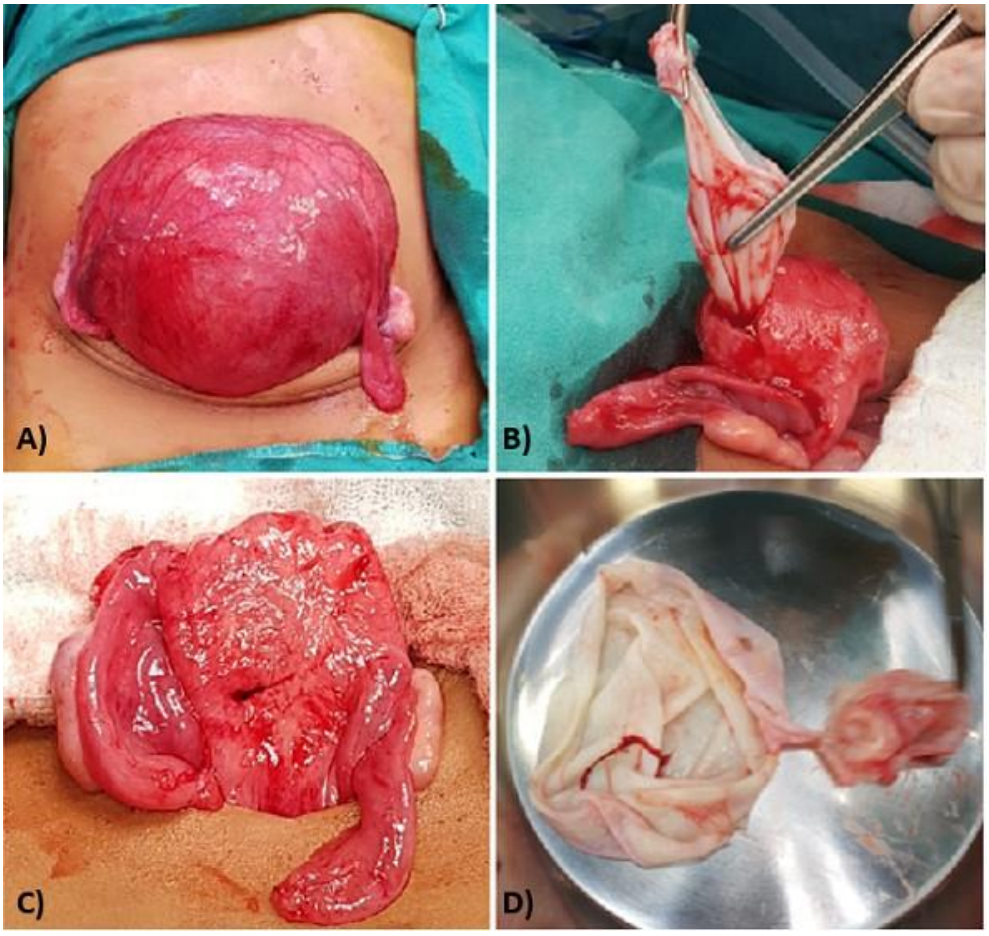

Fig. 2: İntraoperative view of pelvic cyst A) Uterine hydatid cyst revealed by midline incision, B) Removing the germinative membrane from the uterus, C) The appearance of the uterus after cyst removal, D) Germinative membrane

Both ovaries and tuba uterinas were normal. We drained approximately 450 cc of cyst fluid using an infusion set and then introduced 3% saline into the cavity. Then, we completely removed the inner germinal layer of cyst through an incision made in the anterior of the uterine fundus (Fig. 2. B, C and D). Then, we applied total excision to the two cysts in the right and left colon mesentery. Finally, we performed partial cystectomy to the cyst in the liver, and we removed the cyst membrane totally. Drain catheters were placed in the right retrocolic area and pelvis, and the abdomen was closed.

There were no postoperative complication and the patient was discharged on the 7th day. The patient was started on 10 mg/kg/d albendazole treatment after surgery. Histopathological examination was reported as hydatid cyst. The first follow-up 2 weeks later, liver function tests were observed to be normal. The patient continued to be monitored regularly at 3-month intervals. No new cyst formation was observed on ultrasonography. Hydronephrosis completely resolved. The follow-up of the case is at the 26th month and continues uneventful.

Written informed consent was obtained from parent of the patient for the surgery and reporting of this case.

Discussion

Echinococcosis is an endemic disease in many parts of the Middle East as well as other parts of the world including India, Africa, South America, New Zealand, Australia, Southern Europe and Turkey (1,5). Although liver, lung, intraabdominal and spleen involvement is common, pelvic location is extremely rare. Female pelvic organ involvement ranges from 0.2 to 4.27% in the literature (2).

Pelvic organ involvement usually develops secondary to rupture of liver or spleen hydatid cyst (1,3). However, cases of primary uterine hydatid cysts have also been reported (6). In our case, the presence of liver and mesenteric cysts accompanying uterine cyst may support secondary involvement. However, the larger uterine cyst and the lack of knowledge of the prepubertal tubal and fimbrial functions also support the idea of primary location. Therefore, we thought that we could not say with certainty whether uterine involvement is primary or secondary especially for this case.

Hydatid cysts generally tend to grow slowly, and the increase in cyst size has been reported as an average of 0.5 - 3 cm / yr (3). Our case had multiple hydatid cysts, the largest of which was 13 cm in size. Considering this average cyst growth rate, we can say that this parasite was acquired in early childhood. The patient and his family did not notice the distention caused by the uterine cyst in the abdomen. We thought this is due to the slow growth of the cyst. However, hydatid cysts can grow rapidly, albeit rarely. Recently a 5-yr-old case was presented with a 15 cm hydatid cyst in his kidney and accompanying liver cysts (7). The cyst can grow as fast as 3-4 cm/yr (7). We recently presented a 6-yr-old pediatric case with an isolated hydatid cyst around 12 cm in his kidney (8).

Intraabdominal hydatid cysts acquired in childhood may remain asymptomatic until adulthood. In most cases, there are usually no symptoms and it is detected incidentally. However, in cases with pelvic organ cysts, symptoms and signs generally occur secondary to compression of the genital organs, urinary tract, vascular and bone structures (1,5). Dharsandia et al presented a 12-yr-old case with ipsilateral hydronephrosis and renal colic due to right ovarian cyst hydatid (4). Emir et al reported 11-yr-old boy who developed bilateral hydronephrosis and renal failure due to retrovesically located hydatid cyst (9). Retrovesically located hydatid cysts could cause symptoms such as palpable mass, flank pain, pollacuria and abdominal pain. Hydronephrosis in our case was bilateral and it occurred due to the compression of the uterine cyst on the distal ureters and bladder.

The patient became symptomatic with bilateral flank pain that developed secondary to this urinary system compression.

The diagnosis of hydatid cyst is usually made by imaging methods and serology. While positive serology supports the diagnosis, negative results do not exclude the presence of hydatid cyst. Ultrasonography and contrast-enhanced computed tomography are effective imaging methods showing abdominal cysts (6). We performed both of two imaging methods and revealed that there were other cysts in the liver and mesentery concomitant with the pelvic cyst. When a hydatid cyst is detected in any organ in the body, it should be investigated whether there are cysts in other parts of the body.

The choice of treatment method may vary depending on the number, size, and location of cysts and compression of the adjacent organs. If possible, total removal of the cyst is the accepted approach. However, partial cystectomy can be applied in cases where the cyst cannot be removed completely (1). In cases where the cyst is incompletely removed or there is intraoperative spillage, effective results are obtained by early initiation of oral albendazole or mebendazole after surgery (5).

Total hysterectomy is safely applied in cases of uterine or ovary hydatid cysts encountered in the post-menopausal period (2,3). However, preferring this approach in young patients may expose the patient to both organ loss and undesirable complications of hysterectomy. The cyst with an organ-sparing approach was removed in a 27-yr-old young woman with uterine hydatid cyst (6). Similarly, uterine-sparing surgery was performed on a 17-yr-old hydatid cyst case originating from uterine serosa (10). We also preferred uterine-sparing approach because of the fact that our case was still in childhood period. We performed partial cystectomy with only excision of germinative membran. We started oral albendazole treatment in the early postoperative period due to partial excision.

In the literature, there was not enough information about the long-term results of the uterine sparing approach. For this reason, we believe that long-term follow-up of the patient, including the post pubertal period, is essential for future gynecological problems such as uterine synechiae.

Conclusion

In endemic regions, hydatid cysts should be considered for the diagnosis of children with cystic mass lesions. Uterine-sparing approach should be kept in mind as an option, especially in young women. Early surgical treatment of pelvic cysts that cause obstructive uropathy may prevent the progression of renal damage. However, surgical principles should be followed to prevent spilling of cyst contents. Postoperative chemotherapy (albendazole or mebendazole) should follow in cases where complete excision cannot be performed.

Acknowledgements

The authors would like to thank Dr. Mehmet Emin Güldür for his cooperation especially on Parasitologic assessments.

Conflicts of interest

The authors declare that there is no conflict of interest.

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