Case Series

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Retrovesical hydatid cyst: an unusual location of hydatid disease about a case series

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Introduction and importance: Hydatid disease is an endemic zoonosis in regions with temperate climates where pastoral farming is common. Retrovesical localization is rare. Given the rarity of this entity, the lack of personal clinical experience, and the difficulty with detecting early symptoms, the diagnosis remains elusive for years.

Methods: This is a 30-year retrospective, descriptive and analytic study of seven patients who were hospitalized and operated on in the Department of Urology during 30 years (1990–2019).

Outcomes: The average patient age was 54 years (range: 28–76). Signs of bladder irritation were the predominant presenting complaint. No cases of hydaturia were noted. Preoperative diagnosis was based on ultrasonography and serology tests. Hydatid serology was positive for three patients. In three cases, a hydatid cyst of the liver was associated. A partial cystopericystectomy was performed for five patients, it was total for one patient. The resection of the prominent dome was realized once. No cystovesical fistula was found. The mean postoperative stay was 16 days. The postoperative course was uneventful for five patients. Urinary fistula occurred in one patient. One case of infection of the residual cavity was observed. One patient had a retroperitoneal cyst recurrence requiring reoperation.

Conclusion: The preoperative diagnosis of retrovesical hydatid cysts is based mainly on ultrasonography. Open surgery is the treatment of choice. Different approaches are possible. Given the rarity of this entity, management should be guided by experienced experts.

Keywords: cyst, echinococcus, surgery

Introduction

The retrovesical hydatid cyst is an uncommon presentation of this disease even in endemic areas, accounting for approximate accounts for only 0.1–0.5% of hydatid cases and 1–2% in the Tunisian series[1]. The pathogenesis of this entity is explained either by the hematogenous dissemination or the fissuration and subsequent seeding of an intraperitoneal hydatid cyst in the subvesical and retrovesical fat[2]. A direct spread of the embryos through the rectosigmoid mucosa to the pelvis and perivesical venous plexus has also been hypothesized[2].

HIGHLIGHTS

- The retrovesical hydatid cyst is a rare location for hydatid disease.
- Nonspecific symptoms are the main reason for a delayed diagnosis.
- Imaging is the cornerstone of the diagnosis.
- Surgery is the only curative treatment for retrovesical hydatid cysts.
- Surgeons should operate, keeping in mind the existence of adjacent organs (iliac vascular axis, rectum)

Herein we report seven cases of retrovesical hydatid cysts (RVHC) with a special emphasis on diagnostic approach and management modalities.

Patients and methods

It was a retrospective, observational case series study conducted in a single tertiary care center. Institutional Review Board approval was obtained (CEBM. EPS.HCN/62/2022). Our data has been reported in line with the PROCESS (Preferred Reporting Of CasE Series in Surgery) criteria[3]. In this study, the authors confirmed that all methods were carried out under the relevant guidelines and regulations (Helsinki Declaration) under the number research registry 8011. We retrospectively included all patients who were referred to our department for the management of hydatid retrovesical cysts during a 30-year period from

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January 1990 to December 2019. We counted seven consecutive cases. We excluded patients whose medical records were incomplete. A systematic review of patient records and operative reports was performed. The data analyzed were age, personal, medical and surgical history, type of surgery, circumstances of discovery, time of diagnosis, results of biological and radiological examinations, type of surgery, duration of hospitalization, complications, and radiological and biological examinations during monitoring.

Results

Patients ranged in age from 28 to 76 years with an average of 54 years. All seven patients were male. All patients' history documented a rural origin and prolonged close exposure to dogs. No personal history of hydatid disease was found in all cases.

The most frequent complaint was signs of bladder irritation. It occurred in five patients. Three patients presented hypogastric pain. The cyst was discovered incidentally in one patient. No hydaturia nor hematuria was reported. Clinical examination revealed a painless retropubic mass in three patients.

The blood count has shown hypereosinophilia in two patients. The hydatid serology [ELISA (enzyme-linked immunosorbent assay) test] was performed in four cases and was positive in three of them.

Abdominal ultrasonography was performed in all patients. The cystic lesions were classified according to the ultrasonographic classification of Gharbi *et al.*[4]. Type III cyst was observed in three patients. A type I, II, IV, and V cyst was described in one patient each. Five patients had a second hydatid cyst located in another organ. The liver was the most frequent location (three patients), followed by the peritoneum (two patients).

A plain film of the urinary tract was done for four patients. Cystic peripheral linear calcifications were observed in two cases (Fig. 1). A comprehensive intravenous urography was performed for two patients, showing in two cases a mass distorting and displacing of the bladder. A nonfunctional left kidney subsequent to left ureteral compression by a large RVHC was observed in one case (Fig. 2).

The computerized tomography was done for all patients preoperatively to provide additional information regarding the relationship of the cyst to adjacent structures, which is essential for planning the surgical approach and type (Fig. 3). No fissured hydatid cyst was found. The size of the cyst ranged from 80 to 200 mm, with an average of 132 mm (Fig. 4).

Treatment was surgical for all patients. Preoperative antiparasitic oral therapy (albendazole 400 mg/day) was administrated in one patient (concomitant multiple hydatid locations). In one case, preoperative bilateral ureteric stenting was realized, as there were multiple associated peritoneal hydatid cysts to treat in the same session as the RVHC. Surgical access was gained through a midline subumbilical laparotomy in four cases. Three patients underwent large midline laparotomy to allow a total left nephrectomy in one case (nonfunctional kidney) and for multiple associated peritoneal locations in the two other patients. The area around the cyst was then carefully isolated by surgical fields soaked in a scolicidal agent (hypertonic saline or hydrogen peroxide). After sterilization of the cystic content by injection of 10 ml of hypertonic saline (four patients) or hydrogen peroxide (three patients) for 10 min, the cyst was aspirated. The cyst was then opened, and the endocyst was removed (hydatid membrane and daughter cysts). Partial cystopericystectomy with drainage of the residual cavity was performed in five patients. Total cystopericystectomy was realized in one patient, and resection of the prominent dome was performed in one patient. No cystovesical fistula was found. The patients with concomitant hydatid cysts of the liver underwent hepatic surgery in a separate session. No intraoperative or postoperative antiparasitic therapy was used.

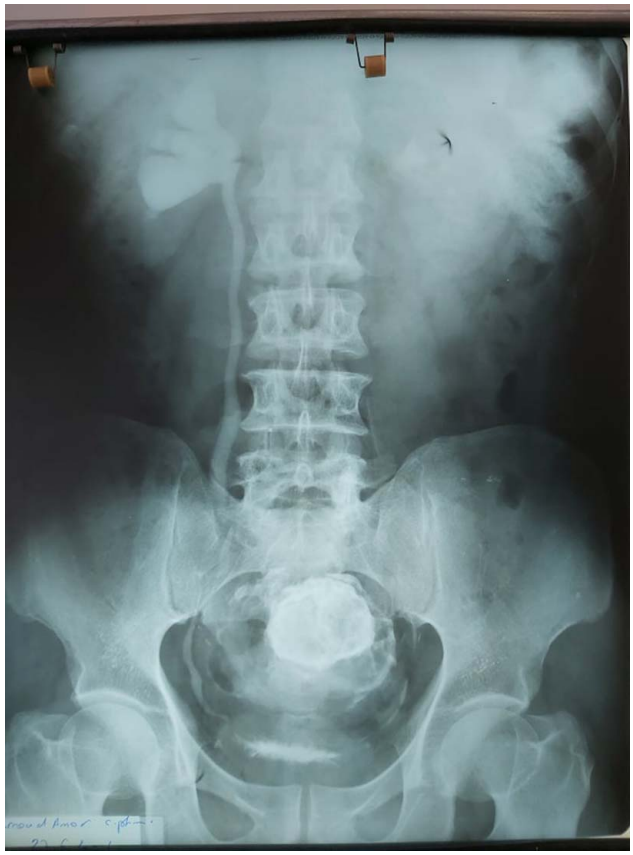

Figure 1. Plain film showing a calcified cyst projecting on the bladder area.

The postoperative course was uneventful for five patients. One patient had urine leakage that ceased after prolonged drainage for 12 days. One patient presented a residual cavity abscess. He had nonsurgical treatment consisting of maintaining the tube drain for 4 weeks.

The hospital stay ranged from 5 days to 46 days, with an average of 16 days. Two patients were lost to follow-up. The remaining five patients were followed for a period varying from 6 weeks to 45 months. A retroperitoneal recurrence was noted in one patient at 9 months, for which he underwent surgery.

Discussion

Hydatid disease is still endemic in Tunisia and the Maghreb countries, where it causes medical, veterinary, and economic problems. In Tunisia, the surgical incidence is 15/100 000[1].

RVHC is rare, with a reported incidence of only 0.1–0.5% of hydatid disease cases[1]. In our urology department, this location

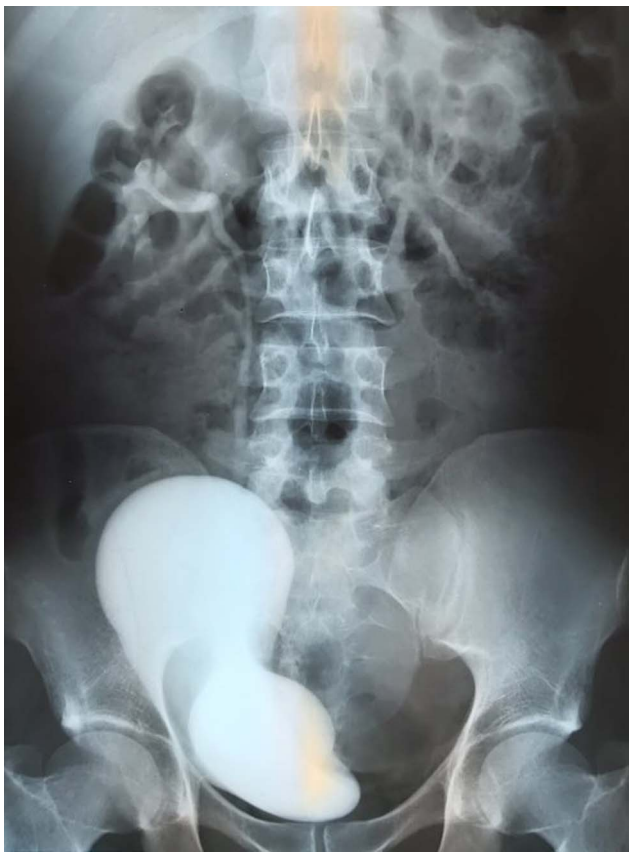

Figure 2. Intravenous urography showing extrinsic compression of the bladder (the left kidney was nonfunctional).

represents 10.7% of the treated urological hydatid cysts during the last 3 decades.

Most RVHCs are considered to be secondary to spontaneous rupture from a primary intra-abdominal location, usually in the liver, or due to inadvertent surgical inoculation, but primary RVHC can occur rarely. RVHC can be considered primary only when no other cysts are present, and in such cases, the hydatid

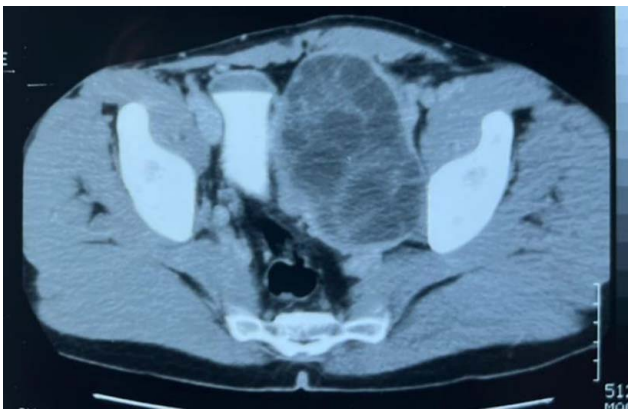

Figure 3. Contrast-enhanced computed tomography scan showing a type III hydatid cyst with compression of the urinary bladder.

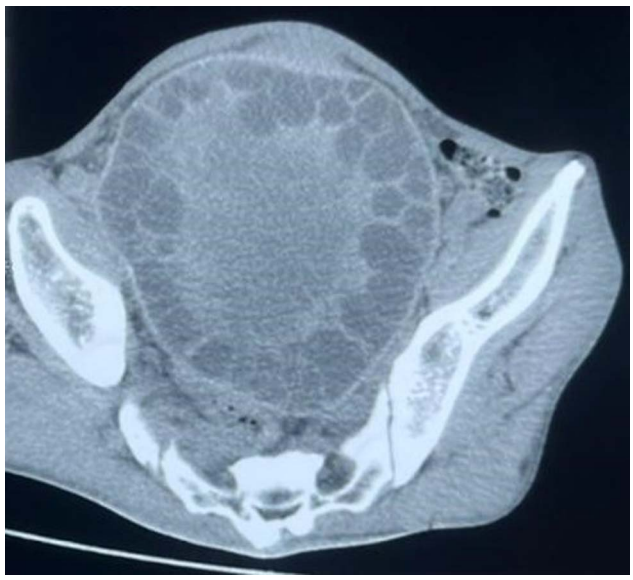

Figure 4. Computed tomography depicting a 200 mm retrovesical hydatid cyst type III of Gharbi.

embryo gains access to the pelvic cavity either by hematogenous dissemination (by passing the hepatic and pulmonary filters) or by the lymphatic route. Direct access of the embryos through the rectosigmoid or vaginal mucosa to the pelvic venous plexuses and perivesical tissues has also been hypothesized[2]. However, the 'secondary echinococcosis' theory cannot always explain isolated RVHCs when a primary intraperitoneal hydatid cyst is absent with no evidence of peritoneal seeding.

Due to its location in a fixed bony pelvic cavity, it usually compresses the bladder, the ureters, and the urethra during its early development[5].

The symptomatology is late-onset due to the insidious evolution of the cyst. The most frequent presenting symptoms are frequency, urgency, burning micturition, and possibly urinary retention. Transit disorders and hemorrhoids, flank, or pelvic pain are also common[2,5]. Seminal vesicle involvement by RVHC can cause hemospermia[6]. A physical examination can reveal a hypogastric firm mass. Three patients of our series presented a retropubic mass on palpation.

Despite the lack of specificity of clinical symptoms or complaints that will reliably confirm the diagnosis of retrovesical hydatid location, hydaturia, although rare, is the only pathognomonic sign of a hydatid cyst ruptured in the urinary tract, and for instance, in the bladder[2]. None of our patients presented hydaturia. Eventually, the cyst is discovered during a complication: suppuration of the cyst, an anaphylactic shock after his fissuration, or a renal insufficiency by ureteral compression[7]. A nonfunctional kidney secondary to ureteral compression was discovered in one patient in our series.

Serology is useful in the diagnosis of hydatid disease. However, at present, there is no serodiagnosis test with a sensitivity and specificity high enough to be totally reliable in the diagnosis of hydatid disease; that is why serology has to be confronted with radiological findings[2].

Intravenous urography can reveal a liquid pelvic mass with distortion and displacement of the bladder. Peripheral calcifications can be present as well as upper tract distension[5].

Ultrasonography, a noninvasive, accessible, and sensitive imaging technique, which is also cost-effective, should be the first-line diagnostic tool. Hydatid cysts may be classified into five types based on the imaging classification of Gharbi *et al.*[4]. Types II, III, and V do not pose a diagnostic problem, but type IV, called pseudotumoral, may lead to confusion: A heterogenous hyperechoic well-circumscribed retrovesical lesion may be evocative of a type IV hydatid cyst, a pelvic abscess, or a pelvic tumor[4]. The presence of other cysts at various stages of evolution in other organs can confirm the diagnosis of a hydatid origin.

In male patients, RVHC may mimic congenital or acquired cysts of the seminal vesicle (also seen in some parasitic diseases such as bilharziasis), posterior bladder diverticulum, the Müllerian duct cyst, the ejaculatory duct cyst or diverticulum, a prostatic cyst, hydronephrosis in the pelvic kidney, a large ureterocele, or an anterior sacral meningocele[5,7]. In female patients, RVHC may be misdiagnosed as an ovarian cyst or tumor, tubal ectopic pregnancy, prolapsed uterine fibroid in the Douglas pouch, or a hydrosalpinx[7].

CT scan is of a great contribution in litigating cases, for instance, pseudotumoral cysts. It depicts a hypodense or hyperdense proper-walled mass that keeps unenhanced with contrast medium. It can also show peripheral calcifications and daughter cysts, which are highly evocative of hydatid origin. The mass can also be filled with a contrast medium if the cyst ruptures in the bladder[7].

Furthermore, CT can provide additional information regarding the organ of origin to assess the repercussion on the upper urinary tract and the relationship of the cyst to adjacent structures, which is essential for planning the surgical approach and technique[8].

CT-guided diagnostic needle is contraindicated when hydatid origin is suspected. This technique carries the risk of dissemination and fatal anaphylactic reaction[5,8].

MRI was assessed in patients with hydatid disease in its various locations and proved to be not cost-effective in the diagnosis of hydatid cysts[9].

All patients with hydatid disease should be treated when diagnosed. Surgery is the mainstay of treatment of all locations of hydatid cysts. It is aimed at managing the cyst and the remaining cavity without spillage or contamination of the operative field.

In order to avoid peritoneal hydatid seeding, secondary suppuration, and postoperative occlusion, primary RVHC with no evidence of intraperitoneal cysts should be managed by an extraperitoneal approach. Secondary RVHCs necessitate a transperitoneal approach that allows the excision of all concomitant hydatid lesions in the same session[2,5]. In our series, two patients were approached transperitoneally to treat other intraperitoneal cysts in the same session. The area around the cyst should be carefully isolated by gauze packs soaked in a scolicidal agent (30% saline solution, hydrogen peroxide, or 1% iodine). The cyst is sterilized by aspiration of the cystic content, followed by the injection of a scolicidal agent for 10 min[5].

Ideally, total cystopericystectomy should be performed; however, when the cyst is adherent to neighboring structures that need to be preserved, such as the rectum and the ureters, partial pericystectomy is an acceptable alternative, sparing plates in contact with adjacent organs[2]. Five of our patients had a partial cystopericystectomy, and only one patient had a total cystopericystectomy. Resection of the prominent dome was performed in one patient because of dense and intimate adherences to the rectum and iliac vessels. Sometimes, preoperative ureteric stenting may be necessary, as in one patient of our series, to prevent ureteral lesions. Closure of a urinary bladder fistula is necessary when present[5].

Drainage of the residual cavity did not protect against superinfection in the case of hydatid cyst retrovesical because the cavity was nondeclive and difficult to drain. If the cyst is infected, it is advisable to install an irrigation drainage system to prevent secondary collections and suppurations of the residual cavity and detect urinary fistula[7].

Preoperative albendazole therapy remains questionable. This treatment should be reserved for limited or disseminated and recurrent cysts or in case of surgical contraindications.

Laparoscopic management of retrovesical cysts with excision of the redundant cyst and pericyst tissue has been rarely reported[10]. The use of the Da Vinci surgical system for this hydatid location was also reported for the first time in 2010[11]. With this technique, it is claimed that the enhanced magnification, three-dimensional vision, and endowrist technology ensure accurate dissection with no collateral damage[11].

The strength of this study is the rarity of the pathology, mainly in this location. Additional reports are always welcomed to help detailing and improve the management of this entity. There were some limitations in our study. First, with the small sample size, which results in a relatively low number of constatations. Second, surgery was only conducted by open surgery.

Conclusion

RVHC is a rare entity, even in highly endemic areas. The diagnosis, sometimes unclear and delayed, is based on imaging. The treatment is surgical. Regular radiological and biological postoperative follow-up is necessary to detect hydatid recurrence. Prevention and early diagnosis are the keys to reducing the incidence and morbidity of this disease.

Ethical approval

The approval of the current study has been granted by the Medical Committee of Research Ethics of Charles Nicolle Hospital. It is available for review by the Editor-in-Chief of this journal on request.

Patient consent

Written informed consent was obtained from the patients for publication of this study. A copy of the written consent is available for review by the Editor on request.

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We do not have any financial sources for our research.

Conflicts of interest disclosure

The authors declare that there are no conflicts of interest to disclose.

Provenance and peer review

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