Case report

Multiple unusual locations of hydatid cysts including bladder, psoas muscle and liver

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Abstract

Our case concerns 66-year-old female with a multiple unusual locations of hydatid cysts including bladder, psoas muscle and liver. Coexistence of hydatid cysts in these localizations has not been previously reported. The diagnosis of vesical hydatid cyst was facilitated by the coexistence of other echinococcosis locations. Treatment consists of the excision of the cysts in the same session without any postoperative anthelmintic drugs. In a two-year follow-up no recurrence has occurred.

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1. Introduction

Hydatidosis is a human disease caused by the larval form of Taenia echinococcus, which lives in the gut of the dog, wild canines and other carnivorous animals, that represents the definitive host and involves both domestic and wild animals. Humans become the accidental intermediate hosts by ingesting Taenia eggs. Echinococcus is endemic in many countries where sheep, dogs and man live in close contact [1]. The sheep–dog cycle is dominant in North Africa, Middle East, India, and the sheep-rearing areas of South America and Australia.

All organs in the human body may be affected by hydatid disease. Excluding liver and lungs all the other organs of the human body are considered as uncommon locations. Urinary tract involvement is not common, corresponding to only 2–4% of cases [2]. Among the unusual presentations of the echinococcal cyst, the localization in the bladder is exceedingly rare.

2. Case report

A 66-year-old female from a rural region of Tunisia, presented with a left flank pain, complaints of abdominal discomfort, urgency and nocturia. On questioning, she denied any gross hematuria or grape skin-like material present in the urine. Physical examination revealed a palpable right hypochondrial mass. Her renal function was normal and there was no significant eosinophilia.

The plain abdominal X-ray revealed right upper quadrant calcification. Hepatic ultrasonography showed a partially calcified hydatid cyst measuring 7 cm in diameter. Bladder ultrasonography revealed a 3 cm, well-circumscribed, round, mural cystic mass with a central hypoechogenic image (Fig. 1). Cystoscopy revealed a mass projecting on the anterior side, but the bladder mucosa was normal. Abdominal Computed tomography showed a 7 cm partially calcified cystic mass at the right lobe of the liver (Fig. 2) and revealed a 4 cm well-circumscribed cyst of the left psoas muscle (Fig. 3). Pelvic Computed tomography showed a thick-walled 3 cm mural cystic water-attenuation mass at the anterior side of the bladder. Cyst wall enhances after injection of contrast material (Fig. 4A and B). Chest X-ray did not show any
abnormality. Indirect hemagglutination test for hydatid disease was negative. All these findings were interpreted as suggestive of multiple organ involvement of a hydatid disease.

The patient underwent single-stage surgery. Via a left iliac incision, the first step of the surgical procedure was excision of the bladder cyst. The cystic mass was exposed using the extraperitoneal approach. It was found arising from the anterior wall of the bladder. It was a mural 3 cm thick-walled cyst. The cyst was excised totally with the surrounding bladder wall. The bladder was drained by a transurethral catheter.

Via the same incision and using an extraperitoneal approach, the second step involved partial excision for the psoas muscle

Fig. 1. US image showing a 3 cm round mass of the bladder wall with a central hypoechoic image.

Fig. 2. Contrast-enhanced CT scan demonstrates a 7 cm, unenhanced hypoattenuation cystic mass that occupies the right lobe of the liver.

Fig. 3. Contrast-enhanced CT scan showing 4 cm well-circumscribed hypoattenuation lesion that originates from the left psoas muscle.

Fig. 4. CT scan demonstrates a vesical mass. A) Unenhanced CT scan through the lower pelvis shows low-attenuation mass at the anterior side of the bladder. B) Contrast-enhanced CT scan demonstrates a cyst of the anterior bladder wall with homogeneous water-density content, and a peripheral enhancing wall that protrudes into the bladder lumen.
operative diagnosis of hydatidosis. However, diagnosis may be
made by visualizing daughter vesicles in cystic lesions, which is important for a pre-
operative examination; cuticular membrane, germinative membrane and
daughter vesicles were seen in histopathologic section of the hydatid cysts. No complications developed during the post-
operative period. The patient recovered completely and was
discharged from the hospital on the 7th postoperative day. After
two years of follow-up with ultrasonography and CT, the patient
was free of symptoms with no evidence of residual or recurrent
disease.

3. Discussion

Hydatid disease is still a major health problem that affects
both humans and animals in Tunisia. The average annual
incidence rate is 11.3 per 100,000 inhabitants [3]. The vast
majority of the patients resides in rural areas. Typically cystic
hydatidosis consists of a single unicellular cyst. However, 30% of
cases may have synchronous multiple cysts in the same or
multiple organs [4]. We report multiple atypical involvement
including bladder, psoas muscle and liver. Coexistence of
hydatid cysts in these localizations has not been previously
reported. This case supports the overall opinion that hydatid
cysts can be located simultaneously in various tissues.

Hydatid disease located in the psoas muscle is very uncommon
[5]. The occurrence of bladder hydatid cysts is exceptional with
only a few case reports found in the literature [4,6–11]. It is
observed approximately in 0.2–0.5% of cases [4,9]. The path-
ogenesis of the bladder hydatid cyst is explained by hematoge-
 nous dissemination with development of the cyst within the
bladder wall. After ingestion of the contaminated food, hexacanth
embryos migrate by portal system to liver where the majority
lodges. Those escaping this hepatic filter pass through the heart to
lungs, and in most intermediate hosts these two sites account for
majority of cysts. Others, however, may return to the heart and be
distributed by systemic circulation to wide range of locations such
the bladder or the psoas muscle [12].

Bladder location is characterized by clinical latency. The cyst
is slow growing and remains asymptomatic for a long period
[1]. Acute urinary retention was an infrequent reported symptom
associated with the bladder hydatid cyst [6,7]. Hydaturia, which
is characterized by the presence in urine of grape skin-like
fragments, is the only preoperative pathognomonic sign and
testifies to the presence of a ruptured hydatid cyst in the bladder.
However, this finding is far from constant. In our case, the
patient presented with urgency, nocturia and symptoms related
to psoas muscle and liver hydatid cyst locations.

The Ultrasonography is highly efficient in detecting germi-
 nal vesicles in cystic lesions, which is important for a pre-
 operative diagnosis of hydatidosis. However, diagnosis may be
difficult due to the unusual localization and the non-typical
image, as the case we report has shown. The diagnosis may be
easier when the lesion has multiple locations involving [9]. CT
is performed in cases with indeterminate sonographic findings
or negative immunological tests. Daughter cysts within a large
cyst and increased density of the hydatid membrane are
pathognomonic signs. A characteristic scan, even in the absence
of positive serology, should be considered diagnostic. CT is also
the best modality for the detection of the calcifications and the
evaluation of all abdominal and pelvic structures [4].

There are different types of serological tests which can be
considered useful for the diagnosis of patients with hydatid disease.
These serological tests may not always be positive, as in this case.
Enzyme-linked immunosorbent assay (ELISA) with use of IgG,
IgA or IgE, latex agglutination, indirect hemagglutination, total
IgE, radioallergosorbent test and enzyme-linked immunoel-
ectrotransfer blot assays are available. Immunoelectrophoresis to
detect specific antibodies against antigens isolated from hydatid
cyst fluid, double diffusion are-5, is specific [13]. In the present
case, indirect hemagglutination was a sub-optimal assay. In
routine laboratory practice, a combination of two different tests at
least is used. Serology has a clear role in the follow-up of patients
after surgery, titers should fall after resection and any subsequent
rise is likely to indicate recurrence.

Surgery is the mainstay of treatment for hydatid cysts. The
principle of surgical therapy is total excision of the cyst or cysts
whenever possible. The surgeon must be careful to remove the
cyst totally avoiding spilling its contents. Laparoscopic ap-
proach appears safe and effective for the hepatic hydatid cysts
[14]. Vaidyanathan et al. [15] reported 2 cases of pelvic hydatid
ysts communicating with the bladder in which this commu-
nication was used advantageously for intravesical instillation of
a scolicidal agent to destroy the germinal layer, with a good
outcome. In the present case, therapeutic difficulty was that we
had to operate three hydatid cyst locations in the same session.

The experience with preoperative and postoperative therapy
with the antihelmintic agents was acquired by studies done on
liver hydatid cysts. Mebendazole and albendazole are a
benzimidazole derivative. These drugs interfere with mechanisms
of glucose absorption through the wall of the parasite [16].
The second phase of multicenter clinical trials of albendazole in
human cystic echinococcosis by WHO showed that albendazole
was generally more effective than mebendazole probably because
of superior absorption into the intestinal tract [17]. Supplementary
chemotherapy with anthelmintics is recommended to reduce the
risk of dissemination during surgery and to prevent recurrence.
Other authors believe it is only useful in cases of surgical inac-
cessibility whenever radical surgical procedures are impractica-
ble. According to the WHO guidelines for the treatment of
hydatid disease, chemotherapy is indicated for inoperable patients
and those with multiple cysts scattered in many organs where
surgery can be ineffective or hazardous [18]. In the case we report,
all the cysts were removed, then we did not require adjuvant
chemotherapy. Patient remained asymptomatic during the follow-
up without recurrence.

In this paper, we described a rare case of multiple hydatid
cyst located in unusual sites. Coexistence of hydatid cysts in the

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bladder, the psoas muscle and the liver has not been previously reported. The diagnosis of vesical hydatid cyst was facilitated by the coexistence of other hydatidosis locations. Hydatid disease should be kept in mind when a cystic lesion is encountered anywhere in the body, mainly in patients from endemic regions.

References