The Tree that Hides the Forest: Pelvic Hydatid Cyst that Reveals Two Localizations: Pulmonary and Hepatic: A Case Reports

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Received: March 05, 2018; Published: March 24, 2018

Abstract

In Algeria hydatidosis is endemic and is mainly found in breeding areas. The liver is the organ most often affected with a percentage of 90% of cases followed by the pulmonary localization. Some localization is rare. The pelvic localization is exceptional and it is postponed only in a collegiate way. The symptomatology remains nonspecific which poses a real preoperative diagnostic problem. Ultrasound and computed tomography coupled with hydatid serology is very contributory to the diagnosis.

We raise in a case recorded in our service of childhood surgery Constantine, a rare localization of supra-vesical pelvic hydatidosis in a patient of 3 years, revealed by a urinary syndrome whose clinical and radiological investigation revealed the presence of another pulmonary localization. We raise the diagnostic and therapeutic difficulties and the peculiarity of the management in the infant child.

Keywords: Child; Urinary Syndrome; Hydatid Cyst; Pelvic Mass; Secondary Localization; Surgery

Introduction

Hepatic and pulmonary localizations are the most frequent in hydatidosis. We report the case of a 3-year-old girl with pararectal supra-vesical pelvic hydatid cyst, a rare and illusory localization associated with dual pulmonary and hepatic localization. The remedy is classic surgical hydatid cyst in this pelvic localization, with peculiarities of own risk of involvement of the neighboring organs and especially of intraoperative tactics, given the difficulty of differential diagnosis. The aspiration puncture followed by sterilization and the removal of the proliferous membrane remains valid, and is therefore the most appropriate therapeutic strategy.

Case Report

She is a 3-year-old female child from an urban background with no family history who presented in the emergency department to pediatric surgery for dysuria. The parents also reported urinary symptomatology of dysuria and pollakiuria; episodes of constipation; without abdominal pain or haemorrhage the general condition was preserved.

Physical examination highlights; a double pelvic mass and an epigastric, hard to palpation, well limited to regular edges, the digital rectal examination combined with the abdominal palpation allows the perception of anterior bulge of the rectal mucosa, with regular surface, located at 3 cm of the anal margin. The biological assessment, namely blood count formula with eosinophil count, CRP, VS, alfa fetoprotein, and HCG beta were without abnormality. Ultrasound as well as the abdominopelvic CT scan reveal a large cystic formation with a thin and regular pelvic-abdominal wall measuring (75 cm x 71 cm) and a width of 43 cm crossed by thin septa, also a cystic formation of the left liver. 60 cm long axis.

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Figure 1: Pelvic ultrasound: heterogeneous hypoechoic mass.

Figure 2: Liver ultrasound liver echogenic mass.

Figure 3: Axial CT scan, after injection of the intra-abdominal cystic mass contrast material.

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Radio chest face performed in search of another location, actually objective a well round opacity with regular limits of left thoracic medio seat. There was no other hydatid localization, including bone.

**Figure 5:** Front chest radiograph, left medio pulmonary opacity.
A trans-thoracic ultrasound comes as a compliment; revealed the presence of a cystic mass of left mid-lobar hydrate tone 4 cm in diameter.

The hydatid serology by ELISA test was positive at 64 IU/ml and the indirect immunofluorescence reaction at 90 IU/ml.

The patient was initially operated for pulmonary localization, for fear of cystic rupture in the airways, a left thoracotomy has allowed the realization of aspiration puncture and sterilization of the residual cavity and peri-cytectomy.

In a second time after three months of the 1st intervention, a right transversal approach was performed in our patient. Discovering an intraperitoneal hydatid cyst located on the anterior surface of the rectum pushing down the bladder and laterally the two ureters. consisting of several stalls separated by septas unrelated to the vascular axes. Another localization at the level of the liver, occupying the entire left liver with diaphragmatic adhesions where it was then performed aspiration-puncture and sterilization of the cyst with serum salty hypertonic at 30%.

Figure 6: Trans-thoracic echography cystic mass of the left lower lobe.

Figure 7: Pelvic cystic mass per-operatively.
The postoperative course was simple.

The patient was asymptomatic one month after the intervention, we received the piece of surgical excision with anatomopathological and parasitological study. Consisting of a proliferous membrane of the hydatid cyst. Microscopic examination of the centrifugation pellet of the hydatid liquid showed scolex and many hooks confirming the diagnosis.

**Discussion**

Hydatidosis is a zoonosis caused by the larval form of Echinococcus granulosus, a parasitic tapeworm, the digestive tract of the dog which constitutes the definitive host secondary to the consumption of parasitized viscera [1]. Sheep, the main reservoir of the tapeworm echinococcus The man represents an epidemiological stalemate, it is therefore only an accidental intermediate host, it is infected either directly in contact with the parasitized dog or indirectly by ingestion of contaminated food [1]. This is the case of our patient who has no context.

The parasite is found on every continent, with areas of high prevalence in China, Central Asia, the Middle East, East Africa and parts of South America. In the region Mediterranean, including Algeria that is endemic with a prevalence of hydatidosis can reach up to 5% of the population of high endemic areas [2].

The most frequent location is the liver (59 to 75%), followed by the lung (27%), the kidneys (3%), the bones (1 to 4%), the brain (1 to 2%). Other locations, such as the heart, pancreas, spleen, omentum, ovaries, parameters, pelvis, thyroid, orbit, retroperitoneum, and muscles are very rare [3].

Pelvic localization is found in 0.3 to 0.27% of cases [1,3]. In this clinical form, contamination is usually peritoneal and is then secondary to intra-abdominal fissure or rupture of a hydatid cyst of the liver or spleen [4]. The primary pelvic form is exceptional, and the contamina-
tion in this case is by blood or lymphatic system from a parasite having passed the hepatic and pulmonary barriers [3]. This is the case of this patient of three years, or the hydatid cyst was intact.

Regarding pathophysiology, the proposed causes of unusual presentation are filter mechanism of the liver and lungs, dissemination through enteric lymphatic channels, and patency of the ductus arteriosus in early infancy of hydatid disease in children [5].

Of variable virulence, ten distinct genotypes (G1-G10) were identified with a potential impact on pathology, age of onset; the duration of evolution; rethinking treatment. The epidemiology of each genotype has been associated with a particular host, differentiated by their pathogenicity to humans, by the duration of the pre-patent period in the definitive host and by the antigenicity. Thus, recent research has revealed that the vaccine antigen (EG95), developed for the genotype G1, currently classification within Echinococcus granulosus sensu lato has been revised. The strains were classified individually or in groups, and new species were defined according to an Australian review [6]. A more recent study to note that there is more than 47 subspecies pathogenic in north of Iraq [7]. The age of three years is unusual and early for contracted hydatid cyst, which was confusing for diagnosis, so it is a genotype of extreme animosity with a notion of rapid evolution which gave a clinical picture so advanced with multiple localization at an early age, the pelvic mass in the girl to leave thought to the ovarian problem, which pushed us to carried out hormonological assays. The cloisonne aspect and the localized pelvic localization close to this found in cystic lympho-myalgia, surgical exploration will make the difference a differential diagnosis that must be kept in mind until the perioperative exploration. The clinical symptomatology is nonspecific, something that has been confirmed by a koweiti study of 51 cases of hydatique disease reported between 1956 and 1960 [8]. The most frequent mode of disclosure is the discovery of a pelvic mass or a compression of neighboring organs that can give urinary signs (dysuria, pollakiuria, hysturia which is pathognomonic and reflects cyst fissure in the bladder, digestive (constipation), gynecological (metrorrhagia), neurological [4].

Our patient had a urinary and digestive symptomatology. The clinical picture is misleading and the interrogation must then look for the risk factors: the rural environment, the notion of animal breeding, contact with dogs, a history of surgery for hepatic hydatidosis [9]. The disease can also be a complication such as superinfection, rupture, obstructive renal failure by compression of the ureters or opening of the cyst in the bladder which remains exceptional [1,3,4].

Imaging is essential for diagnosis and pre-treatment assessment. Ultrasonography is an innocuous first-line examination with a diagnostic reliability estimated at 96%. The ultrasound appearance reproduces the stages of Gharbi's classification and reflects the evolutionary stage of the disease. Imaging remains the main tool for establishing the diagnosis. Type like those described during the hydatid cyst of the liver:

- Type I: pure anechoic image well limited;
- Type II: anechoic image with membrane detachment;
- Type III: multivesicular fluid collection;
- Type IV: mass of heterogeneous echostructure, pseudotumoral;
- Type V: calcified cyst [10].

The scanner provides more concise information about location, size, number, and neighboring organs [2,10].

Our patient had a type III hydatid cyst. The differential diagnosis arises with an ovarian cyst, cystic lymphangioma, digestive duplication, which are perpetual pathologies in this age group.

Types II, III and V do not pose a diagnostic problem, but type IV, called pseudotumoral, is difficult to distinguish from an ovarian mass of a dermoid cyst or abscess [4,6]. The cystic cystic aspect allows limited diagnostic presumptions in our patient to two: hydatid cyst for context and biology and cystic lymphangioma. CT scan then finds its indication in this case by confirming the diagnosis and to search for other localization in the abdominal cavity [4]. Thorax radiography is sufficient to search for a pulmonary localization. The hydatid serol-
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...logy is positive in 30 to 70% of cases for extrahepatic localizations. Eosinophilia is suggestive in 33 to 53% of cases [6]. Our patient did not have eosinophilia but the hydatid serology by ELISA test was positive. Eosinophilia hyper is inconstant and has only interest in the orientation of the diagnosis. Biology is essentially a question of hydatid serology. It is of a great diagnostic contribution when it is positive. Its negativity does not eliminate the diagnosis of hydatid cyst, hence the need for a confrontation between clinical practice, imaging and biology [6]. Our observation illustrates the interest of the hydatid serology in the positive diagnosis of this parasitosis, however, there is a non-negligible proportion of false negatives, variable according to the localization of the cyst [6]. To improve the sensitivity/specificity ratio, most authors prefer to associate two serological techniques, one quantitative: indirect haemagglutination, immunofluorescence, ELISA and the other qualitative: immunoelectrophoresis, electrosynerese [2]. In addition to its role in diagnostic confirmation, the hydatid serology makes it possible to follow the post-therapeutic evolution of the hydatid cyst, to formulate a prognosis and to detect early secondary hydatidosis. Thus, any elevation in the semester following the intervention is synonymous with recurrences or hydatid locations that have gone unnoticed. Western Blot and immunoblotting are more sensitive and more specific [11].

The treatment of the hydatid cyst is surgical [12]. The approach must be wide. Sterilization of the cyst and protection of surgical drapes is recommended by scolicidal solutions (usually hypertonic saline or hydrogen peroxide). It is recommended to perform a cystectomy or peri-cyctectomy if the cyst is accessible [1,3,12]. Partial pericystectomy is another therapeutic modality for deep cysts and in contact with vascular elements. In some cases, partial or complete removal of an organ invaded by hydatidosis may be necessary. The abdominal cavity must be examined in its entirety in search of other localizations including hepatic [4]. The postoperative treatment based on benzimidazole derivatives (albendazole 15 mg/kg) for several months to prevent recurrence was recommended by some teams, something justified in our patient given the multiple location and virulence of the parasite. Postoperative monitoring is based on clinical examination, hydatid serology and imaging [10]. The last clinical check, three months after surgery, was normal.

The peculiarity of this observation, in addition to the pelvic location of the cyst, lies in the treatment adopted by aspiration-puncture, to avoid damage to neighboring organs or anaphylactic reaction following a dispersion of the hydatid fluid in the peritoneal cavity.

Conclusion

Pelvic localization is an exceptional localization of hydatidosis. The clinical symptomatology is often misleading although rare, this diagnosis is to evoke before a pelvic process in endemic countries for hydatidosis.

Molecular biology has upset the management of this pathology, by identifying the exact genotype and the appropriate management. Ultrasound and computed tomography are of great help in the positive diagnosis. The treatment is surgical.

In the pelvic location of the hydatid cyst, a puncture-aspiration should be preferred without surgical removal of the cyst. A pathology with heavy consequences, especially in young children (scoliosis post-thoracotomy, recurrence postoperative flanges).

Due the benefit of promulgation of legislation in which specific measures are included to interrupt the transmission of the parasite.

Involvement of policies, and inter-professional collaboration (between public health, animal health, municipal health offices, populations). Pending the diffusion of the vaccine against hydatid cyst prevention is the only guarantee solution.

Conflict of Interest

The authors have no conflict of interest.

Bibliography


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