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PRIMARY PELVIC HYDATID CYST WITH SCIATIC COMPRESSION

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Hydatid cysts are endemic in certain regions of the world and particulary in North Africa. They are usually located in the liver, lung, and spleen, though many uncommon locations have been reported. This is the first report of a child with primary pelvic hydatid disease causing a sciatic compression.

Key-word: Echinococcosis.

Hydatid disease caused by echinococcus granulosus is endemic in Tunisia where the incidence is 241cases/year (1). It is therefore among common surgical and diagnosis problems in Tunisia. Hydatid disease has a predilection to locate in the liver and lung (90%). It can also be encountered in almost every part of the body from the crown of the head (1) to the big toe (2).

Pelvic localizations represent 1-2% of all locations in the Tunisian publications and 1% in the European ones (3).

We report on one case of pelvic hydatic cyst and expose the clinical and radiological characteristics of the disease and the therapeutic management particular of this rare location.

Case report

An 8- year-old boy with no specific illness was admitted for exploration of sciatica pain and limping lasting for 2 months in a context of weight loss and asthenia.

Abdominal examination showed a painless pelvic non mobile mass without hepatomegaly or splenomegaly. At digital rectal examination regular para-rectal mass without sphincter troubles was detected. The neurological examination showed left sciatica pain, limping and negative Achilles's reflex.

The electromyography pointed to a lesion at the L5-S1 level.

Biological explorations demonstrated normal blood concentration of αfoetoprotein and βHCG were normal.

Chest X ray was normal. Abdominal Ultrasound revealed a 10 x 9 cm cystic pelvic mass without septas and with vegetations circumscribed by a proper wall.

Post gadolinium abdominal MRI (Fig. 1) shows an oval pelvic mass (9 cm) circumscribed by a thin enhancing wall with a mass effect on the bladder and the rectosigmoid colon and compression of the homolateral sciatica.

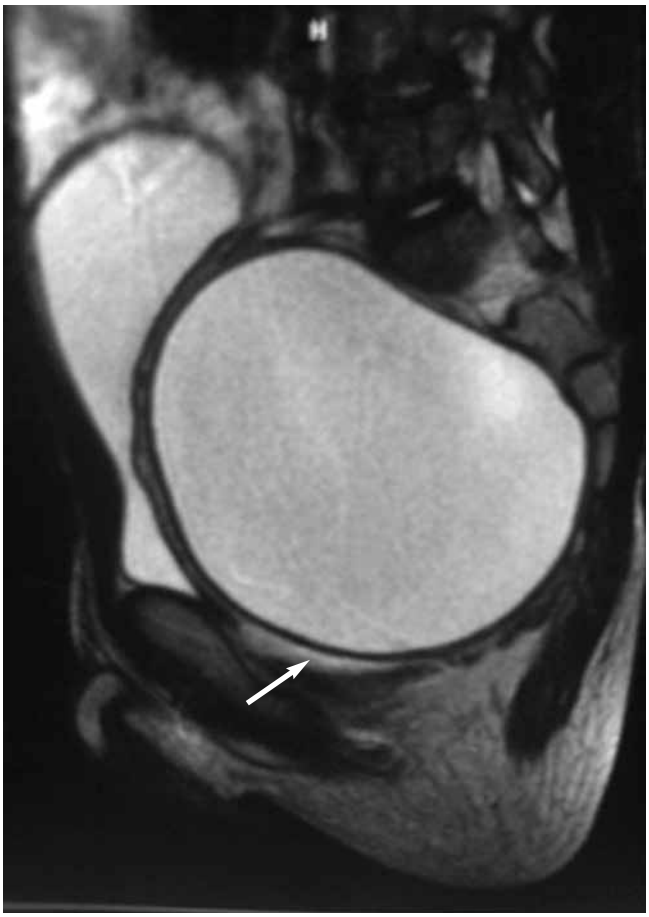

Fig. 1. — Pelvic mass (9 cm) circumscribed by a thin wall pushing back the bladder and rectosigmoid (white arrow).

At surgical exploration (Fig. 2) a retroperitoneal mass pushing the rectum to the right and the iliac vessels to the left was found. The aspect of prominent dome evocated hydatic cyst.

The surrounding structures were protected with gauzes soaked in normal saline solution as a filter against macroscopic spillage. The cyst was then punctured and aspirated, taking extreme care to prevent inadvertent spillage of intracystic fluid. The hypertonic saline (20% NaCl)

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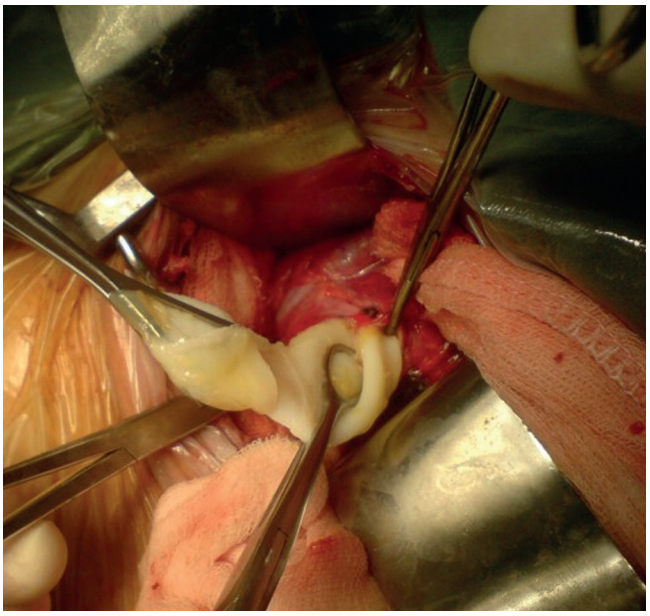

Fig. 2. — The cyst was opened and a discus proligerus was extracted.

solution was injected into the cyst as a scolicidal agent. After 5 minutes, a suction device with side holes was inserted into the cyst and the fluid was evacuated. The germinal membrane was then pulled out. The remaining protuberant pericystic wall was widely excised with insertion of an omental flap into the remaining cystic cavity.

Postoperative recovery was prompt and uneventful with decrease of pain and limping. The patient received albendazole treatment for one year. Follow up after 2 years showed a regression of the neurological symptomatology.

Discussion

Retroperitoneal and retrovesical locations of a hydatid cyst are rare even in endemic areas. According to the theory of Deve, fissuring or rupture of a primary hepatic, splenic or mesenteric cyst would seed its contents in the abdominal cavity (4). This primary cyst might then heal and even disappear, leaving a scar that could be overlooked. The pouch of Douglas would then be the preferred site for the development of a secondary cyst in the pelvis, initially intraperitoneal and later subperitoneal (5).

In the absence of a primary visceral lesion and of peritoneal seeding, hematogenous dissemination could explain the pathogenesis of a solitary retroperitoneal lesion. Oncospheres hatch and penetrate the intestinal wall disseminating primarily to the liver, secondarily to the lung and finally anywhere to form unilocular cysts. They also can pass through the liver and lung barriers, without seeding these structures, and develop an implant anywhere (6, 7). Other pathogenic hypotheses for an isolated retroperitoneal or retrovesical cyst have also been proposed (8, 6): migration of the larvae from the intestinal lymph vessels to the thoracic channel and then anywhere in the body through the hemorrhoidal vessels to achieve a prerectal or retrovesical location or from the rectal ampulla.

Retroperitoneal cysts generally presents as a palpable mass or with flank pain as in our case. A mass was palpable on digital rectal examination. Digestive symptoms, such as constipation, abdominal pain due to the compression effect of the mass are reported. Neurological compression is in theory possible but exceptionally reported. Our patient presented limping and sciatica pain and neurological defect negative Achilles's reflex of the left foot.

The imaging findings frequently suggest hydatid disease but are usually inconclusive and a differential diagnosis may not be made before surgery. A retrovesical cyst mimics many processes, including embryonal cyst, lymphangioma, and digestive duplicity.

Serological tests are undeniable tools for diagnosis and followup of hydatitose. Enzyme-linked immunosorbent assay (ELISA) and immunoelectrophoresis are available. Indirect hemagglutination and ELISA are the most sensitive immunological methods to diagnose human hydatidosis but a false-positive reaction secondary to cross-reactivity with other parasitic infections is possible. In the United States the Centers for Disease Control currently recommends a combination of specific ELISA and Western blot serology (9).

According to the World Health Organization study group on echinococcosis, surgery is still the treatment of choice to provide complete cure.

The optimal treatment of primary retroperitoneal hydatic cyst is complete removal of the cyst without contamination of the field and without sacrificing the organs involved through the appropriate abdominal incision (10).

Removal of germinal epithelium and fluid with scoleces may cause hydatid dissemination and allergic manifestations, even anaphylactic shock. Ideally then, total cyst excision or pericystectomy should be performed (3, 11).

If the localization of the cyst and invasion to vital structures prevent the total excision, partial pericystectomy is the treatment of choice after injection of scolocidal agent followed by removal of germinative membrane (12-14).

Prophylactic measures, such as irrigation with a scolicidal solution are strongly recommended. Hypertonic 30% saline solution for local irrigation, evacuation of the cyst content may be necessary when the hydatid fluid is under high tension. Aspiration of the cyst has been considered an option to standard surgical therapy for elderly patients and an alternative to partial cyst excision or pericystectomy in patients with unresectable disease in the liver. Aspiration of a third of the cyst volume is followed by instillation of the same volume of 95% ethanol within the cyst.

Chowbey et al reported a patient whose RHC was removed endoscopically, but this procedure was applied to only few cases and requires further study (12).

Preoperative treatment with benzimidazoles (albendazole,

mebendazole) has been reported to soften the cyst and to reduce intracystic pressure, enabling the surgeons to remove the endocyst more easily (10). Postoperative treatment with benzimidazoles of patients can reduce the rate of recurrence (10, 13). It is particularly recommended if there is cyst spillage during surgery or partial cyst removal (12). The prevalence of long-term recurrence ranges between 2 and 25%. Recurrence can be due to incomplete cyst removal or previously undetected cysts (10).

Conclusion

In endemic regions, HC should be considered in the differential diagnosis of retroperitoneal cystic lesions. Clinical symptoms are no specific, and appear long time at a late stage of the development of the parasit. Abdomino-pelvic US is the most useful diagnostic tool, and MRI is efficient in the differential diagnosis.

Total cyst excision should be tried in all cases. When this is not possible, removal of all germinative membranes and partial pericystectomy with the use of scolocidal agents are the treatments of choice. Additional adjuvant medical therapy is essential to avoid recurrence.

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