

## CASE REPORT

## Primary Adrenal Hydatid Cyst: A Case Report

**Sataa Sallami, Sami Ben Rhouma, Ali Horchani**

Department of Urology, La rabta Hospital, Tunis, Tunisia

\*Corresponding author: Sataa Sallami Email: sataa\_sallami@yahoo.fr

Published: 01 January 2010

Ibnosina Journal of Medicine and Biomedical Sciences 2010, 2(1):38-41

Received: 06 July 2009

Accepted: 21 November 2009

This article is available from: <http://www.ijmbs.org>

This is an Open Access article distributed under the terms of the Creative Commons Attribution 3.0 License

which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

### Abstract

A 38 year old woman who presented with right flank pain of unclear aetiology. Imaging studies demonstrated a cystic mass in the right upper pole of the kidney causing the ureteropelvic junction obstruction with a significant proximal urinary tract dilatation. She underwent a careful excision of the cyst and the pathological examination confirmed a hydatid cyst (HC) of the right adrenal gland.

We describe the radiological and perioperative findings of this rare case. We have also reviewed the recent medical literature of similar case reports.

**Key words:** adrenal gland, hydatid disease, echinococcosis, hydatid cyst, ureteropelvic junction.

### Introduction

Hydatid disease is an endemic illness in many countries, and it poses an important public health problem. Hydatid disease is a parasitic disease caused by the larval stage of *Echinococcus granulosus* and *E. alveolaris*. Humans may become intermediate hosts through contact with a definitive host (usually a domesticated dog) or ingestion of

contaminated water or vegetables (1).

The adult worm lives in the small intestine of the primary host. Ova are passed in feces and ingested by the intermediate host, which may include humans. Hatched embryos migrate through the intestinal mucosa, enter venules and lymphatics, and reach the liver. If embryos bypass the liver, they can reach the lungs via the inferior vena cava and heart. Embryos may reach any other organ or tissue in the body as the adrenal glands, via the systemic circulation, (2) where it develops into small HC (1,3). Infection with *Echinococcus* larvae generally results in the formation of hydatid cysts in the liver and lungs (2,4), although the infection may affect almost any organ in the body including brain, muscle, kidney, heart and even endocrine glands. Unusual sites for this disease may cause diagnostic problems making correct preoperative diagnosis difficult. The localization of HC in the adrenal gland is extremely rare (5). Only 17 cases have been described in the literature (2,6). With the wider application of ultrasonography (US) and computed tomography (CT) more of these cases are now being reported.

Herein, we report a case of an adrenal hydatid cyst causing uretero-pelvic junction obstruction in a patient with right flank pain.

### Case Report

A 38-year-old woman was referred to us for right flank pain. She denied any hematuria or lower urinary tract symptoms. Her medical history was not significant.

Physical examination was unremarkable. Laboratory data included a complete blood cell count, electrolytes, eosinophil count, serum biochemistry and urinalysis were within normal limits. A plain abdominal X-ray showed a 3-cm round opacity in the right upper region of the abdomen, with curvilinear calcifications (Fig. 1). Abdominal Ultrasound showed a calcified heterogeneous cystic mass



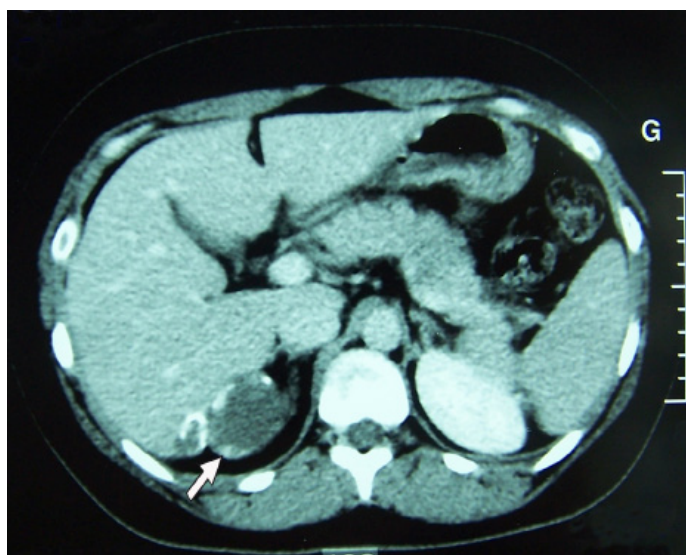
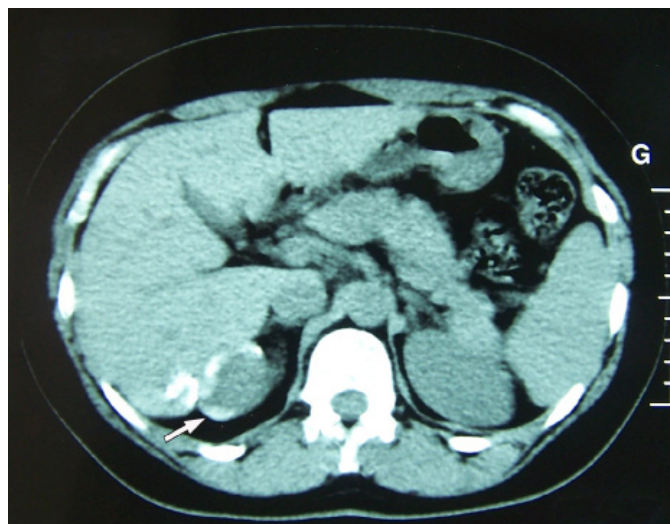
**Figure 1:** Abdominal X-ray: a small round opacity in the right upper region of the abdomen, with curvilinear calcifications (projection on the adrenal gland area) (arrow).



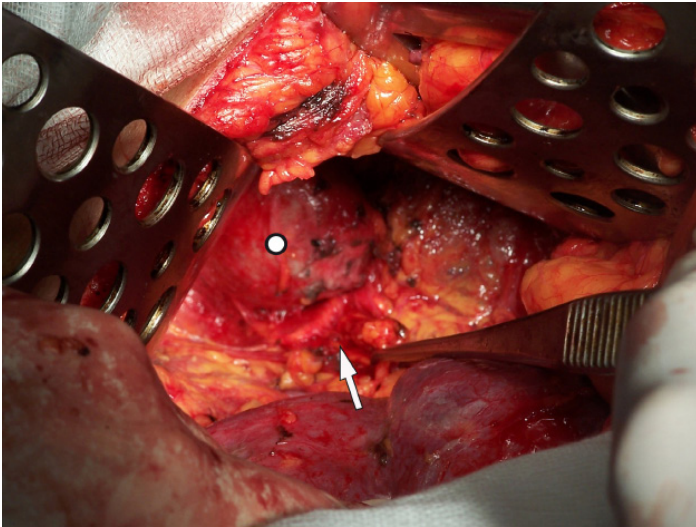
**Figure 2:** IVP: a wall calcified upper renal mass associated with ureteropelvic junction obstruction of the right kidney (arrow).

between the superior pole of right kidney and liver in the retroperitoneal area with pelvic and caliceal dilation. The intravenous pyelography showed the right ureteropelvic junction obstruction with a significant proximal urinary tract dilatation (Fig. 2).

Abdominal computed tomography scan confirmed the ureteropelvic junction obstruction secondary to a solitary cystic mass, located in the upper pole of the kidney. It showed peripheral calcifications, wall thickening, and central partial fluid image with no enhancement after intravenous contrast media injection (Fig. 3, 4). It was 50 x 70 x 50 mm in dimensions. Radiological findings were suggestive of a primary adrenal echinococcosis. No other



**Figure 3, 4:** CT of the abdominal cavity: a cyst in the right retroperitoneal space, above the upper pole of the right kidney with curvilinear calcifications of the wall (arrow)



**Figure 5:** Intraoperative view of the cyst (circle) during the dissection of the adherences with the liver and the adrenal gland (arrow).

intra-abdominal or intra-thoracic masses were found, either in the liver, peritoneum or lung. Chest X-ray was normal. The specific serology for immunoglobulin anti-E granulosus was negative. Urinary catecholamines and metanephrine levels were within normal limits. The patient underwent surgery and a thick-walled 70 mm cyst was dissected out from the surrounding tissues (Fig. 5). At the same session an Anderson-Hynes pyeloplasty with double J stenting was performed via antegrade approach. Inspection of the cyst showed the typical clear fluid of a hydatid cyst with sand pasty material and calcified bodies. Pathological examination of the specimen confirmed a hydatid cyst of the adrenal gland. The patient's postoperative course was uneventful. She was discharged on the third postoperative day. Patient is now six months post surgery and remains well without any symptoms.

### Discussion

Hydatidosis is a parasitic disease (2), endemic in many parts of the world. It is most commonly found in the Middle East, north of Africa, Australia, Iceland and South America, and also remain endemic Tunisia (4). Hydatid disease is capable of involving almost any organ (3). In a series of 275 patients (5), the sites of involvement (in decreasing order of frequency) included the liver (74.8%), lungs (48.3%), peritoneum, kidney, brain, mediastinum, heart, bone, soft tissues, spinal cord, spleen, pleura, adrenal glands, bladder, ovary, scrotum, and thyroid gland. Patients may present with disseminated disease.

Primary hydatid disease of the adrenal gland is a rare event

(4); as this entity has been reported in only 7% of all adrenal cysts and constitutes less than 1% of all cases (5). Adrenal hydatid cysts usually form in association with generalized echinococcosis. Adrenal glands get seeded with infection due to a secondary spread resulting from spontaneous or intraoperative rupture of a primary cyst. On the other hand primary hydatid cysts of the adrenal gland are extremely rare. Out of the 9 adrenal cysts reported by Akçay MN et al, only five of them were primary (5). As the growth of HC is slow, patients generally remain asymptomatic and the diagnosis is made incidentally (5). When symptoms are present, most are related to local visceral compression. The most important clinical features are: flank pain (4,5) and gastrointestinal symptoms (bloating, fullness, nausea, vomiting, constipation, and anorexia) (3). Physical examination is usually normal (4) but occasionally a mass may be found (5,7). The complications of an adrenal gland HC include rupture in the peritoneum or retroperitoneum, local infection, fistula, hemorrhage, or compression of adjacent tissues (5,7). Rarely, an adrenal HC may cause arterial hypertension which has been described as the Goldblatt phenomenon (4,7). Serological studies like ELISA or serum IgE levels may assist in diagnosis (2), but lack both sensitivity and specificity (8). Radiological imaging techniques provide an important aid in the diagnosis and follow up of echinococcosis (5,7). Imaging findings depend on the stage of cyst growth and associated complications (9). In 2003, the World Health Organization proposed the following classification based on ultrasound(US) features of the cysts: type 1 is a well-defined, anechoic lesion; type 2 demonstrates separation of the membrane (the "water lily" sign formed by the undulating membrane); type 3 is characterized by the presence of septa and intraluminal daughter cysts. Type 4 is a nonspecific solid mass. Type 5 is a solid mass with a calcified capsule (9). The case described in this report is a type 5 cyst. A CAT scan is superior to US in demonstrating cyst wall calcification, infection, and peritoneal seeding. It also better delineates the relationship of the cyst with adjacent organs (2). Magnetic resonance (MR) imaging shows the characteristic low signal intensity rim of the hydatid cyst on T2- weighted images (10). Both MR and CT images are able to show the exact anatomic extent, size, volume and position of the mass, the number of cysts, the relationship to other organs and possible complications. Few reports suggest MRI to be more specific than CT (10). Calcification is seen on radiography in 20%-30% of hydatid cysts as a peripheral curvilinear or ring-like pattern. Complete calcification of the cysts is suggestive of the parasite death(6). The presence of calcifications in the

adrenal mass very much supports a diagnosis of HC (8). The preoperative diagnosis of HC is highly suggested by US and CT scan findings (4) but the hydatid nature of the cyst can only be confirmed pathologically.

The differential diagnosis of an adrenal cyst include an endothelial cyst, a pseudocyst due to infarction or hemorrhage, a cystic neoplasm like lymphangioma, a post-traumatic cyst (5,8), a cystic pheochromocytoma (4), an abscess and other congenital or acquired cysts. Just as in hepatic and pulmonary hydatidosis, surgery remains the mainstay for the treatment of HC (5,7). Surgery with either partial or total excision of the cyst, with or without preservation of the adrenal gland, is the treatment of choice (4).

The rapid development of laparoscopic techniques has encouraged surgeons to replicate principles of conventional hydatid surgery using a minimally invasive approach. Among the several suggested surgical methods, the simple resection of the cyst is considered the best option as it allows preservation of the gland. As reported by Horchani and Dionigi (2, 4) open or laparoscopic surgery may be indicated. Secondary infestation can be prevented by intraoperative injection of concentrated saline into the cyst, which causes death of the parasites.

### Conclusion

Primary hydatid cysts in the adrenal gland are a rare Occurrence. Imaging modalities such as US, CT and/or MRI may be helpful in suggesting a diagnosis but confirmation requires a biopsy. Radiologists and surgeons should consider HD in the differential diagnosis of cystic lesions especially in patients from endemic regions.

### References

1. Lewall DB. Hydatid disease: biology, pathology, imaging and classification. *Clin Radiol* 1998;53:863-874 Polat P, Kantarci M, Alper F, Suma S, Koruyucu MB, Okur A. Hydatid disease from head to toe. *Radiographics*. 2003;23:475-94.
2. Dingier G, Carrafiello G, Recaldini C, Sessa F, Boni L, Rovera F, et al. Laparoscopic resection of a primary hydatid cyst of the adrenal gland: a case report. *J Med Case Reports*. 2007;1:61.
3. Polat P, Kantarci M, Alper F, Suma S, Koruyucu M, Okur A. Hydatid disease from head to toe. *Radiographics*. 2003;23:475-94.
4. Horchani A, Nouira Y, Nouira K, Bedioui H, Menif E, Safta ZB. Hydatid cyst of the adrenal gland: a clinical study of six cases. *ScientificWorldJournal* .

- 2006 21; 6: 2420-5.
5. Akcay MN, Akcay G, Balik AA, Boyuk A: Hydatid cysts of the adrenal gland: review of nine patients. *World J Surg*. 2004;28:97-9.
6. Ruiz-Rabelo JF, Gomez-Alvarez M, Sanchez-Rodriguez J, Rufian S. Complications of extrahepatic echinococcosis: Fistulization of an adrenal hydatid cyst into the intestine. *World J Gastroenterol*. 2008; 14:1467-9.
7. Escudero MD, Sabater L, Calvete J, Camps B, Labios M, Lledo S: Arterial hypertension due to primary adrenal hydatid cyst. *Surgery*. 2002;132: 894-5.
8. Otal P, Escourrou G, Mazerolles C, Janne d'Othee B, Mezghani S, Musso S, Colombier D, Rousseau H, Joffre F. Imaging features of uncommon adrenal masses with histopathologic correlation. *Radiographics*. 1999;19:569-81.
9. WHO Informal Working Group: International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings. *Acta Trop*. 2003;85:253-61.
10. Marani SA, Canossi GC, Nicoli FA, Alberti GP, Monni SG, Casolo PM. Hydatid disease: MR imaging study. *Radiology*. 1990;175:701-6.
11. Arif S, Bari S, Wani N, Zargar SA, Wani M, Tabassum R, et al. Albendazole as an adjuvant to the standard surgical management of hydatid cyst liver. *Int J Surg*. 2008;6(6):448-51.