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Case Report

Isolated hydatid cyst of adrenal gland with hypertension mimicking Conn's syndrome: a very rare case

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ABSTRACT

Hydatid cyst of the adrenal gland is one of the rare conditions caused by the larval stage of *Echinococcus granulosus*. The incidence of adrenal gland involvement is less than 1% of all hydatid disease in humans and isolated adrenal involvement is extremely rare. Hydatid disease is frequent in endemic regions and sheep farming areas with equal sex distribution. Here, a case of 23 year old female with isolated adrenal gland hydatid cyst is presented, that was evaluated clinically, investigated radiographicaly and by blood investigations and finally histopathology confirmed the diagnosis. No complications occurred at peri and postoperative period. The patient was given 6 cycles of albendazole (10 mg/kg in two divided doses), each for a period of four weeks followed by a week's rest. The patient is on regular follow-up without recurrence in last 1 year.

Keywords: Hydatid cyst, Adrenalectomy, Laparoscopy

INTRODUCTION

Hydatid disease is an endemic illness caused by parasitic infestation of *Echinococcus granulosus* larva. Humans are the intermediate host and dogs being the definitive host. It occurs due to ingestion of contaminated food and water.[1] Liver (45-75%) is the most common site of hydatid cyst followed by lungs (10- 50%).[2] Brain, heart, kidneys and adrenals are unusual sites.[3] Adrenal gland involvement is very rare and isolated adrenal involvement is extremely rare and constitutes less than 1% of cases.[4] The disease is endemic in India, Africa, Australia, South America, New Zealand. Gender distribution is equal in endemic areas.[5] Most of the cases documented in literature were discovered incidentally, very few have been documented to coexist with arterial hypertension.[6]

We herein report a case of isolated adrenal hydatid cyst associated with arterial hypertension mimicking Conn's syndrome.

CASE REPORT

A 23 year old female presented to outpatient department with complains of off and on headache and muscle spasm of bilateral upper and lower limb once in 15-20 days for past one year. Patient was found to be hypertensive. Routine blood investigations were done. She was found to be hypokalemic (K^+ - 1.95 mmol/L) with raised levels of serum aldosterone. Ratio of aldosterone to plasma rennin activity was higher than cut of value. 24 hours urine VMA was within normal limit. A provisional diagnosis of Conn's syndrome was made. CT scan of abdomen revealed a well defined hypodense lesion in the left suprarenal region, likely adrenal in origin (Figure 1). Surgical planning was done and the lesion was approached by transperitoneal laparoscopy. Large adrenal cyst with residual adrenal gland was found, rest of peritoneal cavity was normal. Complete adrenalectomy along with removal of cyst performed and specimen removed in endosac bag. On cut surface a unilocular cyst was identified, showing few areas of whitish deposits

along with few areas of haemorrhage. Microscopic sections showed laminated hyaline membrane of hydatid cyst (Figure 2 and 3). Pericyst comprises of fibrosis and part of normal looking adrenal gland with no evidence of dysplasia. The diagnosis of Adrenal hydatid cyst was made. Serological test for *Echinococcus granulosus* was also positive. Post operatively patient was given 6 cycles of albendazole (10 mg/kg in two divided doses), each for a period of four weeks followed by a week's rest. The patient was under strict follow-up and no recurrence has been documented yet in last 1 year and is relieved of symptoms.

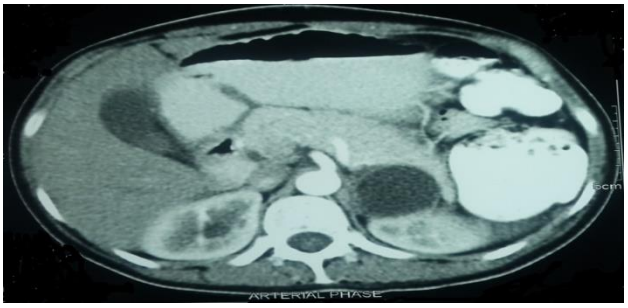

Figure 1: Hypodense lesion in left suprarenal location.

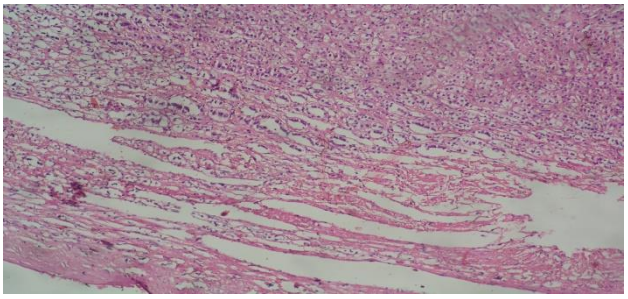

Figure 2: Pericyst with part of adrenal gland; H & E 4 X, 10X).

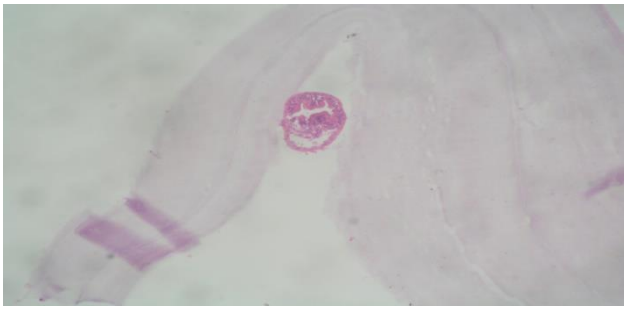

Figure 3: Laminated hyaline membrane of hydatid cyst with broods capsule; H & E 40 X, 10 X.

DISCUSSION

Hydatid disease is a parasitic infestion, endemic in many parts of world like India, Africa, Australia, South America, New Zealand. Gender distribution is equal in endemic areas.[5] Hydatid cyst constitutes only 7% of adrenal cyst.[4] Akcay et al in their study documented 9 patients to have hydatid cyst out of 15 cases of adrenal cysts.[4] Primary hydatid cyst is rare entity.[7] Hydatid cyst of adrenals are slow growing, patients are asymptomatic and diagnosis is incidental.[4] Visceral compression is responsible for symptoms which include flank pain, abdominal fullness, anorexia.[4,7] Goldblatt phenomenon is the association of hypertension with adrenal hydatid cyst.[7,8] This phenomenon is because of irritation of functional tissue by the growing cyst or by renal artery compression.[9,10] It has been reported that even after complete resection of the cyst, hypertension continues.[11] In a case reported by Nouria et al of adrenal hydatid cyst with symptoms mimicking of pheochromocytoma and elevated VMA levels, elevation could be due to adrenal medulla compression by the cyst.[6] In our case urine VMA was within normal limits. Rupture of cyst, local infection, fistula, compression of adjascent tissue, anaphylactic shock is some of the complications.[2] Serological tests aids in diagnosis but they lack in sensitivity and specificity.[12] Imaging modalities plays key role in diagnosing and in follow up.[4] A classification system was proposed by WHO in 2003 based on sonographic features of cysts: type 1 well defined lesion, type 2 demonstrates "water lily sign", type 3 is presence of septa and intraluminal daughter cysts, type 4 is nonspecific solid mass and type 5 is solid mass with calcified capsule.[13] Endothelial cyst, pseudocyst secondary to haemorrhage, lymphangioma, post traumatic cyst, abscess, pheochromocytoma are some of the differential diagnosis.[7] CT guided aspiration of cyst is not recommended because of high risk of dissemination and anaphylactic shock. Treatment is mainly surgical. Total excision of cyst is advisable. Total adrenalectomy is not recommended and is done in those cases where the cyst has completely damaged the gland.[14] Laparoscopic adrenalectomy can also be done and successful management of genitourinary hydatid cyst by laparoscopy has been reported.[15] In our case laparoscopic total adrenalectomy along with cyst excision was performed successfully. Medical management with Albendazole is reserved for in-operable cases and in patients with dissemination.[12]

In our case after histopathological confirmation, imaging studies failed to show any other organ involvement. Thus diagnosis of primary hydatid cyst with arterial hypertension was made.

CONCLUSION

Isolated adrenal hydatid cyst is of rare occurrence. CT scan, USG and MRI are the radiological modalities that are helpful in giving a differential diagnosis. Histopathology is the confirmatory test. It should be considered in the differential diagnosis of patients presenting from endemic regions.

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