J. R. Coll. Surg. Edinb., **43**, October 1998, 347—352

Case report

Some rare presentations of hydatid cyst (*Echinococcus granulosus*)

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Among all the cases of hydatid disease seen in an 8-year period at Asir Central Hospital, Abha, Saudi Arabia, seven cases are reported here because of their unusual presentations. One patient had a brain hydatid cyst which presented as a space-occupying lesion. The second patient presented with symptoms and signs of cardiac tamponade due to pericardial hydatidosis. The third female had multiple abdominal and pelvic hydatid cysts causing vague abdominal pain, chronic ill-health and primary infertility. The fourth case was a huge single hydatid cyst filling the whole abdominal cavity and involving multiple organs. The fifth case presented with simultaneous involvement of the liver, right diaphragm and pleura with hydatidosis. The sixth case involved the left diaphragm and the patient presented with clinical picture simulating pleurisy. The last patient presented with a hydatid cyst of the right thigh. Even though there was no mortality in these patients, there was disabling morbidity. We conclude that *Echinococcus granulosus* can affect any organ in the body and a high suspicion of this disease is justified in endemic regions. Moreover, medical treatment should precede and follow the surgical intervention.

Keywords: abdominal, brain, hydatid cyst, pelvic, pericardium, thigh.

Hydatid disease (*Echinococcus granulosus*) is endemic in the Middle East as well as other parts of the world, including India, Africa, South America, New Zealand, Australia, Turkey and Southern Europe.[1–3] Infestation by hydatid disease in humans most commonly occurs in the liver (55–70%) followed by the lung (18–35%); the two organs can be affected simultaneously in about 5–13% of cases.[4,5] Even though hydatid cysts can occur in any organ, it is very rare to see the disease in the organs reported in this communication. Considerable morbidity and even mortality can be caused by this benign disease, as seen in this report and the literature reviewed.

The purpose of this paper is to emphasize the fact that this disease should be suspected in cystic lesions affecting any organ in the body, especially in endemic areas of the world.

CASE 1

A 12-year-old Saudi female presented with a 4-month-history of progressive left temporo-occipital headache and diminution of vision in the right eye. The right pupil was 4 mm and the left was 5 mm in diameter. The two fundi showed papilloedema. A computed tomography (CT) scan of the brain showed a large cystic lesion in the left parieto-occipital region with air fluid level and midline shift to the right (Figure 1). Exploration through a left parieto-temporal craniotomy revealed a hydatid cyst in the left parietal lobe which was enucleated. Hydatidosis was confirmed histopathologically. Medical treatment was commenced post-operatively and she was discharged in a satisfactory condition 12 days later.

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CASE 2

A 50-year-old Saudi female presented with a 2-month history of severe exertional dyspnoea accompanied by a dry cough. Physical examination revealed a patient with respiratory distress, blood pressure (BP) of 90/60, pulse rate of 100/min and jugular venous pressure (JVP) of 7 cm of H_2O. There were fine crepitations in both lung bases, an 8 cm palpable liver and mild peripheral leg oedema. Total leukocyte count (TLC) was 6700 cells uL^{-1} (3% cosinophils). A chest radiograph showed cardiomegaly, and an echocardiogram showed a huge pericardial effusion which was thought to be tuberculous in origin. After initial resuscitation, left thoracotomy and pericardiotomy were performed through which about 1500 cc of hydatid fluid with daughter cysts were drained. The cavity was irrigated with 0.5% silver nitrate solution. Two pericardial windows were created and pericardial and chest tubes were inserted. Histopathology of the pericardium removed confirmed *Echinococcus granulosus*. Post-operatively, she received medical treatment and was discharged in a satisfactory condition 19 days later.

Eight months later, the patient was re-admitted with dyspnoea on exertion and palpitations. Investigations showed Echinococcus antibody titre of 1:512 and chest radiographical findings were consistent with left ventricular aneurysm or recurrent hydatid cyst. Electrocardiogram (ECG) findings were consistent with lateral wall infarct and an old anterior septal wall infarct. The Echocardiogram confirmed apical aneurysm with laminated thrombus in the left ventricle. The CT scan of the abdomen showed a 6 cm diameter hydatid cyst in the right lobe of the liver which had recently appeared. No surgical intervention was considered necessary at this time. The patient was anticoagulated with Warfarin and put on Albendazole in the hope of medically treating the liver hydatid cyst and discharged in a satisfactory condition.

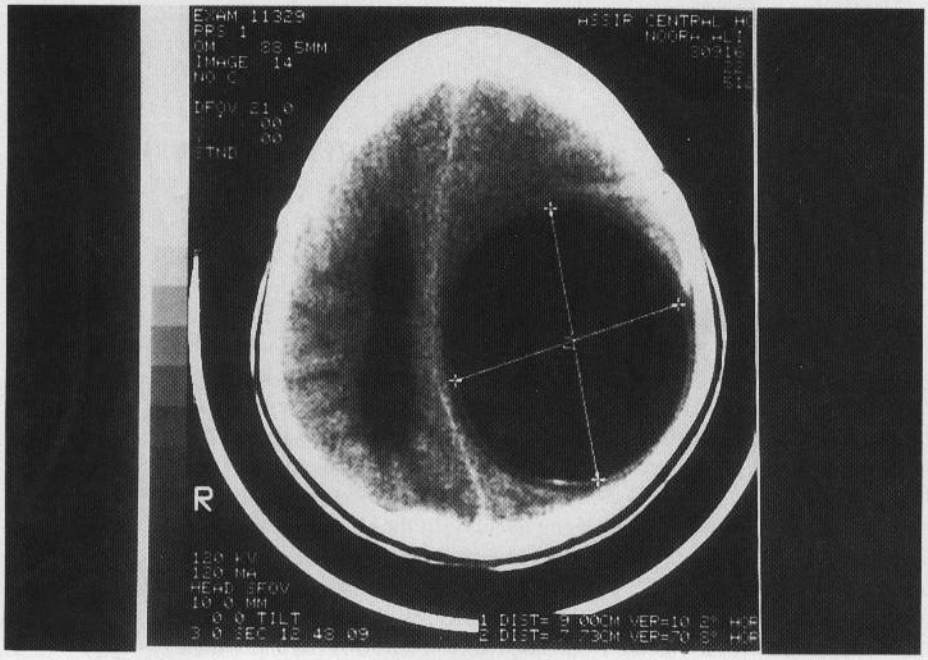

Figure 1 CT scan of the brain showing a large hydatid cyst in the left parieto-temporal region.

CASE 3

A 25-year-old Saudi female presented with a 5-year history of vague abdominal pain and generalized malaise. She underwent laparotomy 10 years previously for a hydatid cyst of the liver. She had been married for the past 10 years and had primary infertility. Clinical examination revealed a pale and cachectic female with numerous, non tender firm masses in the left hypochondrium, left lumbar region, left iliac fossa and in the supra pubic area. She had a palpable liver of about 5 cm below the costal margin. Rectal examination revealed globular masses in the pouch of Douglas. The TLC was 4900 cells uL^{-1} (1% eosinophils). Serological test for hydatidosis was 1:4096 and the abdominal ultrasonography (USS) and CT scan showed multiple cystic masses in the lower pole of the spleen (Figure 2). The right lobe of the liver and the left kidney had been replaced by a lobulated hypodense mass (Figure 3). The left ureter was dilated and draining into another mass in the left iliac fossa (Figure 4). Another similar cystic mass was seen posterior to the uterus. Intravenous urography (IVU) revealed a non-functioning left kidney.

At laparotomy, hydatid cysts of various sizes (2–9 cm in diameters) were found in the spleen, left kidney (multiple), lower end

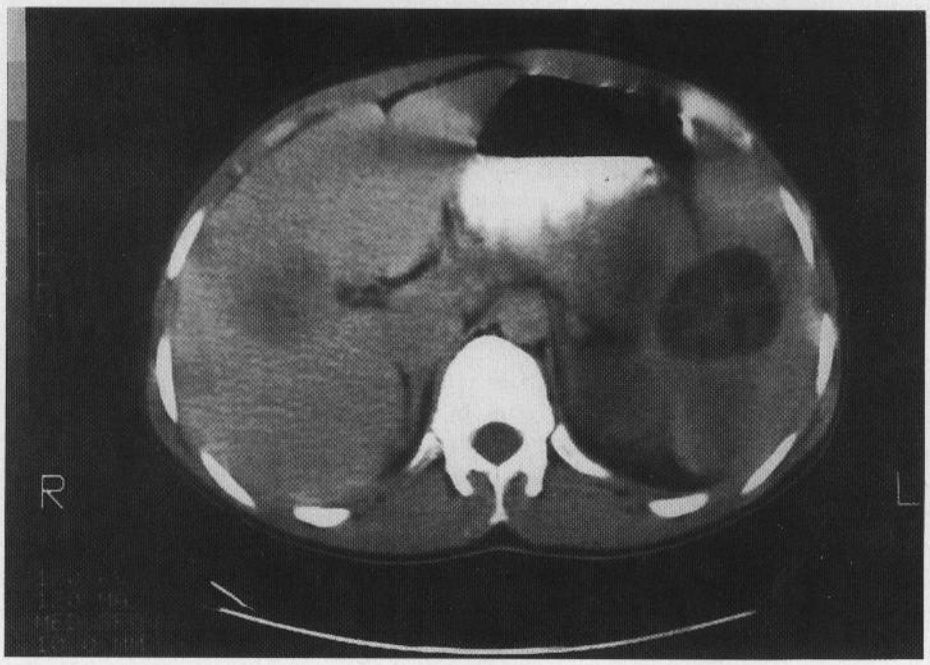

Figure 2 CT scan showing a hydatid cyst in the lower pole of the spleen.

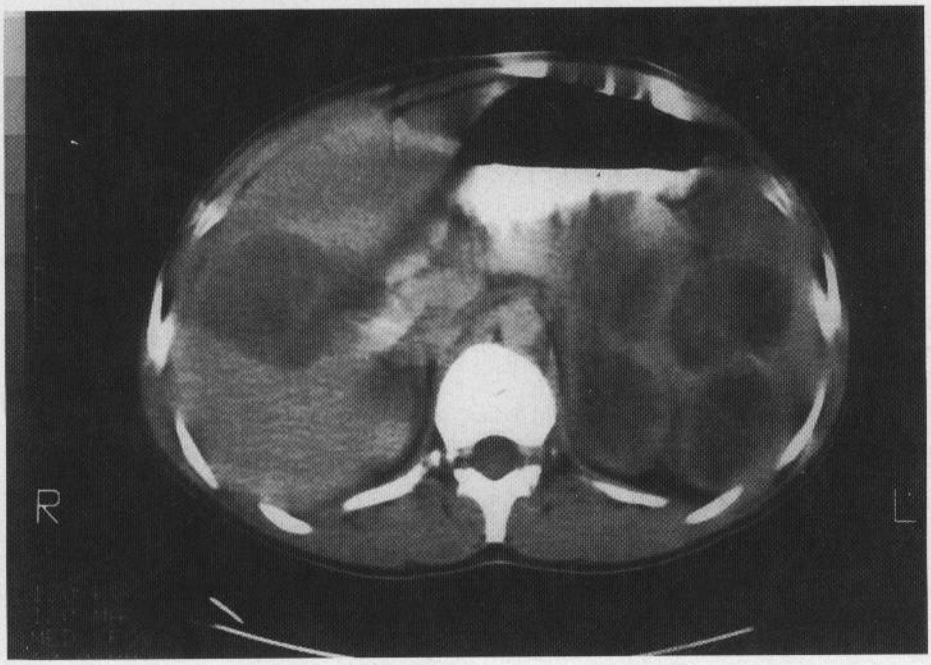

Figure 3 CT scan showing hydatid cysts in the liver and left kidney.

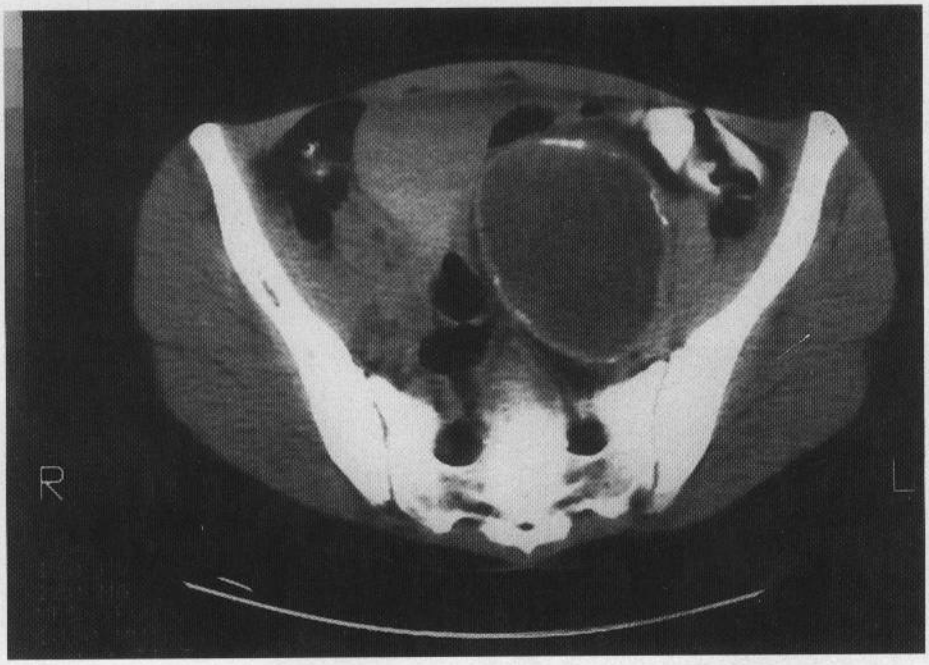

Figure 4 CT scan showing a hydatid cyst in the left iliac fossa.

of the left ureter causing a severe degree of pyonephrosis, pouch of Douglas, the fimbriated end of the right Fallopian tube, and the right lobe of the liver. The uterus was plastered down to both Fallopian tubes which were dilated and full of adhesions. Splenectomy, left nephrectomy, ectocystectomy of the hanging liver cyst and endocystectomy for the rest of the cysts were performed. Histopathology confirmed hydatidosis in all the specimens. Postoperatively she continued on medical treatment, which was started 2 weeks pre-operatively and discharged in a good condition 10 days later.

CASE 4

A 45-year-old Saudi female was admitted with a progressively enlarging abdominal mass of 15-years duration. This had been

associated with abdominal discomfort. Six days prior to admission, she had severe abdominal pain associated with vomiting. Physical examination revealed a thin female with a non-tender, irregular, mobile abdominal mass, partly solid and partly cystic, filling the whole abdomen. The TLC was 5600 cells uL^{-1} (3% Eosinophils). Ultrasonography and a CT scan of the abdomen revealed a large multicystic abdominal mass extending to the pelvis (Figure 5).

At laparotomy, a huge hydatid cyst of about 30 cm in diameter extending from the epigastrium to the pelvis was found. This mass pushed the stomach cephalad, and invaded the spleen and the pancreas. Splenectomy was performed, and the hydatid cyst which was mostly located in the mesentery was excised intact. Histopathological examination confirmed hydatidosis. The patient was put on medical treatment post-operatively and discharged in a satisfactory condition.

CASE 5

A 55-year-old Saudi male presented with severe right hypochondrial pain, nausea, vomiting, fever, cough and chest pain of one day duration. Physical examination revealed tachypnoea, tachycardia and a temperature of 39 °C. He had rigors and chills. Chest examination showed dullness, diminished air entry and bronchial breathing in the right lower zone of the chest posteriorly. There were tenderness and guarding in the right upper quadrant of the abdomen and the right lower intercostal spaces. The TLC was 24700 cells uL^{-1} (0% eosinophils). A chest radiograph showed consolidation of the right lower lobe of the lung with pleural effusion. Abdominal ultrasound was suggestive of two infected hydatid cysts in the liver. On the day of admission, the abdomen was explored through a right subcostal incision. Two intrahepatic hydatid cysts were found with the larger containing thick purulent material. Endocystectomy, after irrigation with 0.5% silver nitrate solution, was performed followed by omentoplasty for these cysts. Two days later, his chest condition did not improve and a chest radiograph showed hydrothorax in the right base. Needle-aspiration revealed pus and after insertion of a chest tube, pus and hydatid daughter cysts were drained. Right posterolateral thoracotomy and drainage of the ruptured hydatid cyst of the pleural cavity were carried out. A smaller cyst in the right diaphragm was also seen. The pleural cavity and the diaphragmatic cyst were washed with 0.5% silver nitrate solution. Decortication of the right lung was performed together with curettage of the hydatid cyst cavity. A chest tube was then inserted. Post-operatively, the patient was put on medical treatment and was discharged in a satisfactory condition.

CASE 6

A 49-year-old Syrian male was admitted twice because of a history of severe, stabbing retrosternal pain radiating to the left shoulder. He had a splenectomy 6 years previously for splenic hydatidosis. Physical examination revealed a young male in respiratory distress and a temperature of 37.8 °C. The left side of the chest was dull to percussion and auscultation showed vesicular breathing in the same area. He was initially thought to have pleurisy with some degree of pneumonia. An echocardiogram revealed a large cystic lesion adjacent to the left ventricle but quite separate from the pericardium. The TLC was 10400 cells uL^{-1} (6% eosinophils). A chest radiograph showed raised left cupola of the diaphragm with atelectasis. Ultrasonography showed a large cystic mass of about $15 \times 10 \times 10 cm^3$ in the left subdiaphragmatic region pushing the left kidney caudally. Serology for hydatid cyst showed a positive titre of 1:512.

At laparotomy, a large hydatid cyst was found below the left crus of the diaphragm and another one at the upper pole of the left kidney. The left crus of the diaphragm formed part of the cyst wall. The cyst was injected with 0.5% silver nitrate solution, and endocystectomy was then performed. The left hemidiaphragm, from where the cyst was excised, was repaired in two layers. The other small cyst in the upper pole of the left kidney was also injected with 0.5% silver nitrate solution, drained and then de-roofed. The

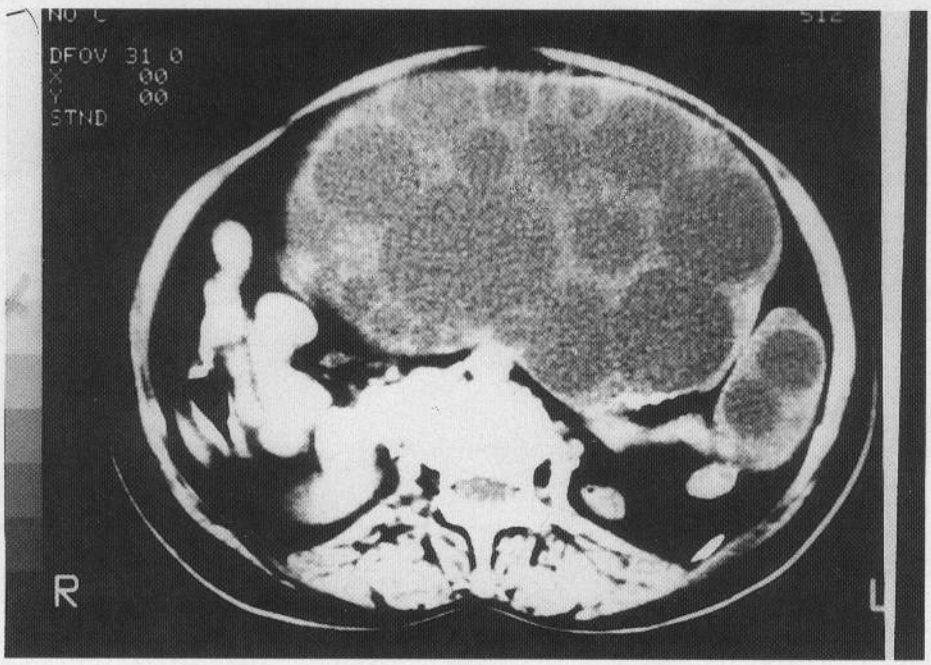

Figure 5 CT scan showing a large multicystic abdominal mass involving the spleen and pancreas.

patient was put on medical treatment and discharged in a satisfactory condition.

CASE 7

A 28-year-old Saudi female was admitted complaining of a painful swelling in the right thigh for 8 months which started gradually to increase in size. There was no history of trauma, fever or weight loss. Physical examination revealed a diffuse, non tender mass in the anterolateral aspect of the right thigh which was attached to the deep muscles. The TLC was 5300 cells uL^{-1} (8% eosinophils). A plain radiograph showed a large soft tissue mass in the proximal anterior aspects of the right thigh with intact bone (Figure 6).

Upon exploration, a hydatid cyst attached to the deep fascia was found. This was excised and the soft tissue was washed with 0.5% silver nitrate solution. Histopathology confirmed echinococcosis. Post-operative abdominal USS was negative for any intra-abdominal hydatid cyst. The patient was put on medical treatment post-operatively and discharged in a satisfactory condition.

DISCUSSION

Hydatid disease due to *Echinococcus granulosus* is endemic in cattle- and sheep-raising regions of the world such as Central Europe, the Mediterranean countries, the Middle East, South America, Australia, New Zealand, and South Africa.[1–3] Although hydatid cysts are known commonly to affect the liver and lung, our experience with this series shows that it can also affect the brain, heart, kidney, ureter, spleen, uterus, fallopian tube, mesentery, pancreas, diaphragm, and muscles. Brain involvement, which is more commonly seen in children, is encountered in 1–2% of the patients and the cysts are usually solitary and have an intraparenchymal localization.[2] Cardiac involvement with echinococcosis is uncommon (0.02%-2%); the left ventricular wall is the most frequent site, but the interventricular septum, right ventricle and left or right atrium may also be involved with varying degrees of complications.[6] Major complications of cardiac hydatid disease result from rupture of the cyst either into the heart or pericardium and death may occur subsequent to anaphylactic shock, cardiac tamponade and systemic or pulmonary hypertension.[7,8] Pancreatic involvement has been reported in 0.25–0.75% of adult cases and the mode of infestation is presumed to be haematogenous, although local spread via the pancreatic or bile ducts has been suggested, as well as peripancreatic lymphatic invasion.[3] Pre-operative diagnosis of hydatid cysts of the pancreas may be difficult, as seen in case 4, because it may be confused with pseudopancreatic, cystadenocarcinoma and true congenital and post-traumatic pancreatic cysts.[9]

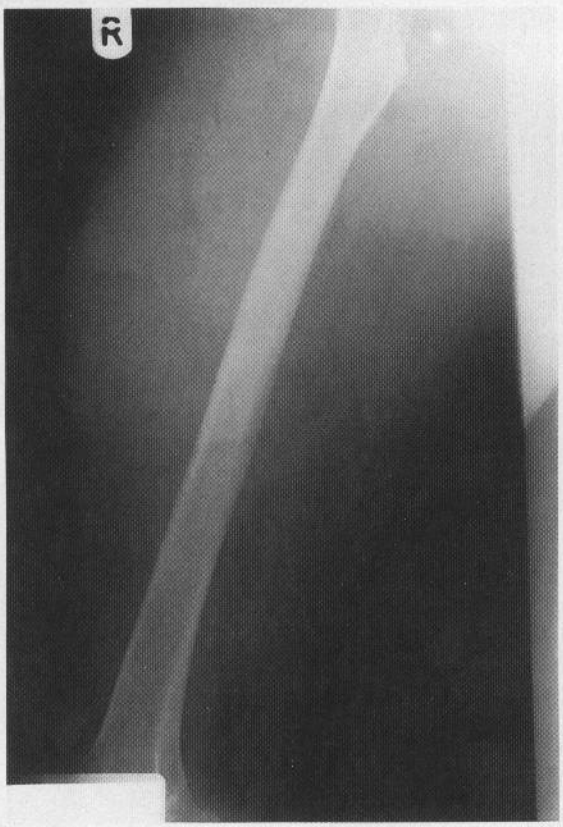

Figure 6 A plain radiograph of the right thigh showing a large soft tissue mass.

The clinical presentation of hydatid disease depends on the size and site of the lesion and the accessibility of the organ involved for clinical examination. Table 1 summarizes the different modes of presentation in our cases. Although eosinophilia is expected in patients with parasitic infestations, this was only seen in two of the reported cases (Table 1). Pre-operative diagnosis of hydatid cysts can be made ultrasonically and confirmed by a CT scan.[3] In this report, as well as others, USS and CT scans were the most helpful investigations[10] (Table 1). Furthermore, echocardiography and magnetic resonance imaging (MRI) are of great value in diagnosing and determining the anatomic extent and relationship of the cyst in cardiac hydatidosis.[6,8] The MRI is also of considerable value in cases of intracranial hydatidosis.[2,11] Different serological tests are being carried out for the diagnosis, screening and post-operative follow up for recurrence. These include the hydatid immunoelectrophoresis, enzyme-linked immunosorbent assay (ELISA), latex agglutination and indirect haemagglutination (IHA) test.[12]

The treatment of hydatid cysts is principally surgical. However, pre- and post-operative 1-month courses of Albendazole and 2 weeks of Praziquantel should be considered in order to sterilize the cyst, decrease the chance of anaphylaxis, decrease the tension in the cyst wall (thus reducing the risk of spillage during surgery) and to reduce the recurrence rate post-operatively.[1,12] Intra-operatively, the use of hypertonic saline or 0.5% silver nitrate solutions before opening the cavities tends to kill the daughter cysts and therefore prevent further spread or anaphylactic reaction.[12] The recurrence encountered in case 3 was most likely due to dissemination from previous surgery.

Even though mortality directly due to echinococcosis is very low, it can produce a very disabling morbidity. A mortality rate between 0.29 and 0.6% has been reported.[13] The recurrence rate of this disease is still relatively high accounting for about 10%.[12] Three out of seven of the patients in this series presented with recurrence of the disease.

We conclude that *Echinococcus granulosus* can affect any organ in the body and a high suspicion of this disease is justified in any cystic neoplasm of any organ, especially in endemic regions. Moreover, medical treatment should precede and follow the surgical intervention.

Table 1 Showing the different modes of presentations and preoperative investigations

Case No.	Mode of presentation	Eosinophils %	Pre-operative suspicion of hydatid disease
1	Headache and diminution of vision	Not done	Yes by CT scan
2	Dyspnoea	3%	No
3	Abdominal pain and infertility	1%	Yes by the past history and CT scan
4	Abdominal swelling	3%	Yes by CT Scan
5	Septicaemia	0%	Yes by USS
6	Retrosternal pain	6%	Yes by the past history and USS
7	Thigh lump	8%	No

ACKNOWLEDGEMENTS

I would like to thank the surgeons whose cases were included in this series and Dr Ismat Riyaz Khalid, FRCSI who assisted in collecting the data for this paper. I also thank Prof. O. G. Ajao, FACS, Prof. Ahmed Ibrahim, FRCS, for their suggestions and review of this paper and Mr Vher Crisostomo for preparing the manuscript.

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Paper accepted on 29 July 1997