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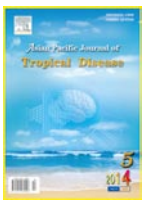

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Reports of four cyst hydatid cases in different size and location

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PEER REVIEW

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Comments

A case series on a tropical disease that can be publishable in the journal. No new knowledge but it can be a record in the database for the follower to cite. The importance can be seen in the field of tropical medicine.
Details on Page 411

ABSTRACT

Hydatid disease/echinococcus is caused by *Echinococcus granulosus* and it is endemic in many countries. Echinococcus commonly affect the liver but it has propensity to involve any organ or unusal location of the body. Treatment is surgical and medical but in many cases desicion of teatment is hard because of the location and size of cyst hydatid. In the present report, authors presented four patients with echinococcus with different anatomical site, size and desicion of treatment.

KEYWORDS

Hydatid cyst, Localization, Therapy

1. Introduction

Echinococcus/hydatidosis is a frequently encountered serious health problem in countries where agriculture and animal husbandry is common. It is frequently seen especially in the Mediterranean region, Australia, South America, Middle East, South Africa, and Eastern Europe[1-3]. It is associated with cystic lesions commonly observed in the liver (50%-80%) and lung (5%-30%), and more rarely in bone, brain, and other organs[1,4,5]. Clinical presentation of the disease varies according to anatomic involvement. While it is usually diagnosed easily, the difficulty of treating the disease depends on the organs involved. In these case reports, authors aim to present four rare cases of hydatid cyst with rare organ involvements and clinical features.

2. Case report

2.1. Case 1

A 38-year-old male patient was admitted with severe back and right hip pain lasting for twelve years. He underwent bone biopsy to exclude malignancy, which was compatible with hydatid cyst. He underwent sacral debridement which was followed by albendazole treatment. For the past ten years, he was intermittently given albendazole treatment and two sacral debridements were performed. However, the pain increased gradually and the patient became immobile. The abdominal computerized tomography (CT) scan revealed 10 cm of multiloculated cysts extending from the right S1 and S2 vertebrae corpus to the right iliac wing. The magnetic resonance imaging (MRI) showed deformity in

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the right coxofemoral joint and right acetabulum (Figure 1). The lesion involved right sacroiliac joint and right iliopsoas muscle. A cutaneous fistula tract was seen in the same region. The patient underwent bone biopsy which revealed hyaline membranes compatible with hydatid cyst (Figure 2). On admission to our clinics, the patient had a lesion with pus drainage on the right hip which was 4 cm in diameter. His erythrocyte sedimentation rate was 128 mm/h and C-reactive protein level was 103 mg/L. He was consulted with orthopedics and neurosurgery departments and he was regarded as inoperable at that time. Because of the extension of the lesions despite albendazole treatment, 25 mg/(kg·day) praziquantel was added to his treatment regimen. On the fifth day of praziquantel treatment, the size of the lesion on the sacral region and pus drainage decreased remarkably. Pus drainage cultures did not show any microorganisms. On the second week of praziquantel treatment, the C-reactive protein level and erythrocyte sedimentation rate decreased to 63 mg/L and 60 mm/h, respectively. Patient was discharged from the hospital with oral albendazole and praziquantel treatment.

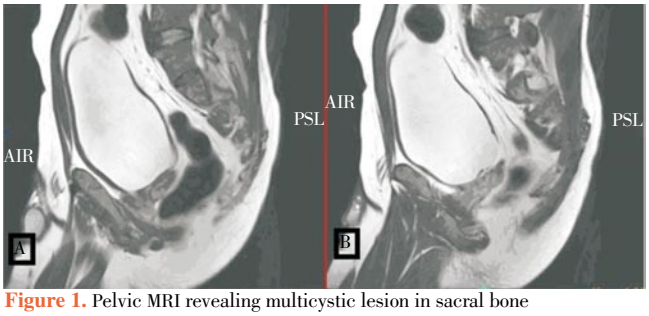

Figure 1. Pelvic MRI revealing multicystic lesion in sacral bone

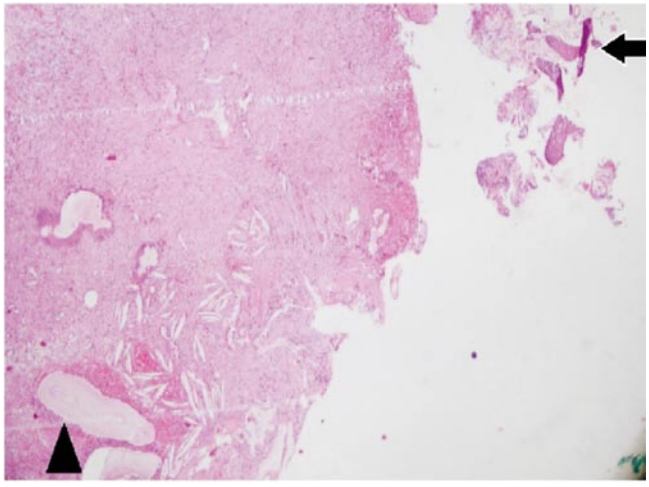

Figure 2. Hydatid cyst germinative membrane.

2.2. Case 2

A 35-year-old male patient admitted to our hospital with complaints of chest pain, shortness of breath and headache. He was living in rural areas. Physical examination revealed right upper quadrant pain on deep palpation. Chest radiography showed either fluid collection or a cystic lesion 10 cm×14 cm in size in the basal right lung. The thoracic CT revealed a bilobed, thick-walled and dense cyst with 14 cm ×9 mm in size between pleural layers in the lower lobe of the right lung (Figure 3). Due to his complaint of headache, he underwent cranial MRI which showed a solid mass or a cyst 3 cm×2 cm in size with parietal dense contrast enhancement in the right frontal lobe and approximately 1 cm subfalcine shift to the left ventricle (Figure 4A). Echinococcus indirect haemagglutination (IHA) test was positive in 1/128 titer. The patient underwent total cystectomy for the right lung cyst. The solid mass in the cranium was supposed to be compatible with hydatid cyst and the left one was unoperated as it was difficult to access due to its localization. He was given 2×400 mg/day albendazole treatment postoperatively and the cyst in the cranium showed significant regression in size to 1 cm ×1 cm (Figure 4B). The patient was decided to be unoperated due to significant improvement in the cranial cyst and discharged with albendezol treatment.

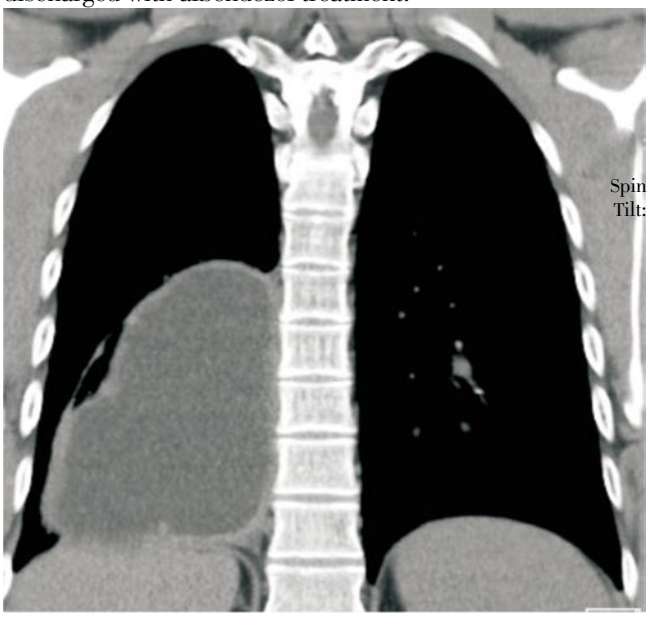

Figure 3. Chest CT showing cystic lesion in the right lung.

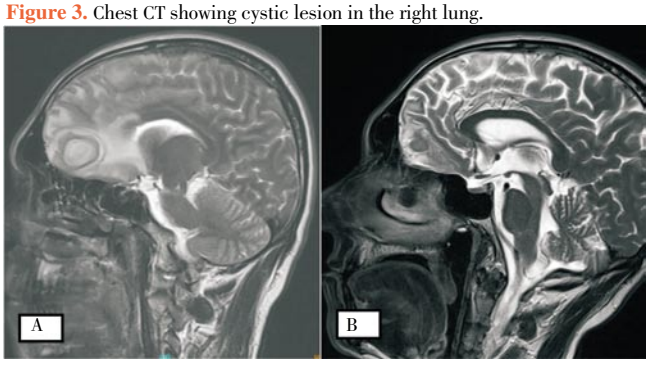

Figure 4. (A): Cranial MRI showing cystic lesion in the right frontal lobe; (B): significant regression of the cystic lesion in the right frontal lobe after two months of albendazole treatment.

2.3. Case 3

A 34-year-old male patient was a guard in a factory and was working with dogs. He admitted to our hospital with complaints of diffuse abdominal pain. The abdominal MRI showed a large T1 hypointense, T2-hyperintense cyst of 181 mm×160 mm invading the whole left lobe of the liver with daughter cysts within and a 129 cm×108 cm× 102 mm sized cyst in the anterior segment and a 107 cm× 95 cm×57 mm sized cyst inferiorly in the right lobe of the liver with multiple septation and daughter cysts (Figure 5). Echinococcus IHA antigen was positive in 1/4096 titer.

The patient was given 2×400 mg albendazol treatment and underwent surgery. However, the cyst could not be removed completely because of the location and size, and he was given albendazol treatment postoperatively.

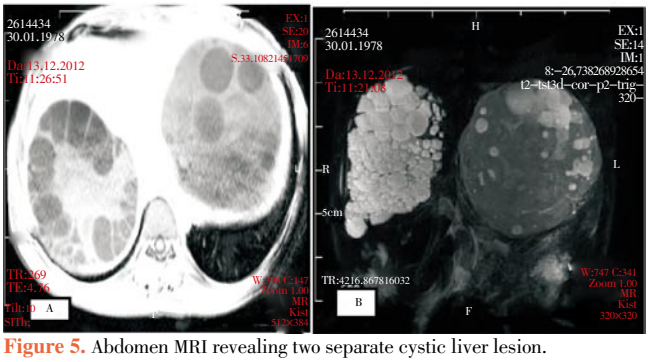

Figure 5. Abdomen MRI revealing two separate cystic liver lesion.

2.4. Case 4

A 40-year-old male patient was admitted with complaints of pain in the right thigh. He had been suffering from the pain for ten years, but the pain gradually worsened in the last year. The MRI of the right thigh revealed 8 cm×2 cm sized multiple cysts located in the muscle tissue adjacent to the corpus of femur (Figure 6). Echinococcus IHA antigen was positive in 1/512 titer. The patient underwent surgery and discharged with albendazol treatment.

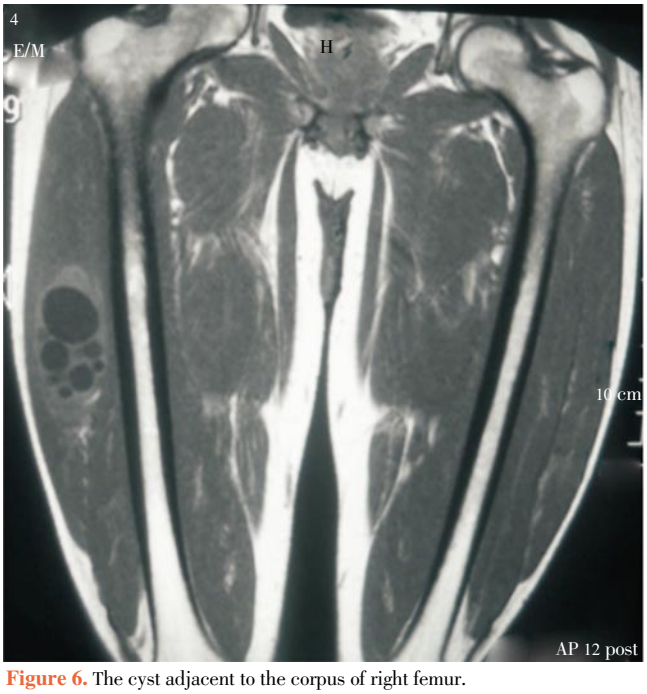

Figure 6. The cyst adjacent to the corpus of right femur.

3. Discussion

Echinococcus granulosus and *Echinococcus multilocularis* cause hydatid disease in humans[6]. The clinical presentations of the echinococcus vary depending on the involvement of the organ and the size of the cysts. While small or calcified cysts may be asymptomatic, large cysts may exert pressure or may rupture[1,7]. The cysts may grow 1-5 cm in size per year, or may stay silent for years. The liver is infected in 2/3 of the cases while lung is the second frequently involved organ[8]. In the presence of liver involvement, cholestatic jaundice, abdominal pain, weight loss, anaphylactic reaction and pancreatitis may occur[9]. In our third case, the cyst caused fatigue and abdominal pain due to compression. The surgical excision of the cyst together with its capsule decreases the rate of recurrence. However, in the third case the capsule could not be removed due to the location of the cyst. For this reason, the patient was discharged with close follow-up and treatment because of the risk of post-operative recurrence.

Pulmonary involvement may cause chronic cough, chest pain, shortness of breath, and hemoptysis[10]. Intrathoracic extrapulmonary hydatid cyst cases are very rare (5%-7%) [11-13]. Dakak *et al*. showed that most of the intrathoracic extrapulmonary hydatid cyst cases involved mediastinum (42%), chest wall (27%), diaphragm (24%) and rarely pleural space (6%)[14]. On the contrary, there are also report showing higher incidence (72.7%) of pleural hydatid cyst cases[12]. In the second case, the hydatid cyst localized in the pleural space and enucleation was achieved.

Cerebral hydatid cyst is rare and accounts for 2%-3% of the cases. Clinical signs of the cyst depend on the localization, size and effect to the organs. Neurological symptoms usually develop due to increased intracranial pressure. The most common initial symptoms are headache and vomiting. Papilledema, hemiparesis, seizures, slurred speech, cranial nerve palsies, and ataxia may also be seen[15]. Lesions are usually single. The cysts are seen the most frequently in supratentorial parietal region of the middle cerebral artery[16]. Cerebral hydatid cysts are divided into two as primary and secondary. The primary type is more common, occurring due to embryos escaping from the body's filter system, and showing fertile and solitary properties. The secondary type is more rare, which occurs due to the embolisation of the scolex after the rupture of the cyst and is usually multiple and infertile. It is most commonly caused from the left ventricle of heart and is therefore referred as metastatic[17]. Surgical removal of the cyst is the most effective method in the treatment of hydatid cyst. Medical treatment is given before and after surgery to prevent postoperative recurrence. In the second case, due to difficulty of surgical removal of the cerebral cyst, the patient followed with medical therapy and 50% regression was achieved with albendazole treatment. Albendazole treatment was continued for the patient with close follow-up with cranial imaging.

In a study from Serbia, bone involvement was seen in 5.6% of all cases[18]. In our first case, isolated sacral involvement was present and neither surgical nor medical therapy was successful. One of the most important problems of hydatid cyst is recurrence of the disease. Combination of the surgical and medical treatment is suggested to prevent recurrences Neelapa *et al*. reported a case of recurrence despite the combination of medical and surgical treatment[19]. Prousalidis *et al*. had reported a recurrency incidence of 8.7%[20].

Muscle involvement is very rare in hydatid disease. In one study, 22 hydatid disease cases with soft tissue involvement

living in endemic rural areas were presented[21]. The thighs (27%) and the gluteal region (9%) are usually involved.

Hydatid disease is still a serious public health problem in our country. It may be seen in different clinical presentations. Therefore, in patients suffering muscle and joint pain and peripheral neurologic symptoms, especially if there is a history of living in rural areas, spinal or pelvic hydatid cyst should be suspected and usual organ involvements such as liver and lung should also be screened. In addition, combination of long-term medical treatment with an effective surgery and careful postoperative follow-up may be useful in prevention of recurrences.

Conflict of interest statement

We declare that we have no conflict of interest.

Comments

Background

Echinococcus/Hidatisosus is a frequently encountered serious health problem in countries where agriculture and animal husbandry is common. It is associated with cystic lesions commonly observed in the liver and lung, and more rarely in bone, brain, and other organs.

Research frontiers

The present report presents four rare cases of hydatid cyst with rare organ involvements and clinical features.

Related reports

There are plenty of reports by other researchers to show the involved organs and incidence of this disease in other regions.

Innovations & breakthroughs

The present case repor listed 4 rare cases of hydatid disease with rare involved organs and clinical presentations, and treatments for the patients were provided as well.

Applications

The present case report on rare cases of hydatid cyst, which is an important disease, can be further referenced.

Peer review

A case series on a tropical disease that can be publishable in the journal. No new knowledge but it can be a record in the database for the follower to cite. The importance can be seen in the field of tropical medicine.

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