CASE REPORTS

Disappearance of Recurrent Intra-abdominal Extrahepatic Hydatid Cyst Following Oral Albendazole Administration

OP Michail, C Georgiou, PO Michail, E Felekouras, I Karavokyros, G Marinos, A Giannopoulos, J Griniatsos

ABSTRACT

A 44-year old male patient with a past medical history of a complete surgical excision of pelvic hydatid cyst two years previously presented with constant pelvic floor pain and plasma IgG anti-echinococcal antibody titres of 14.27 U/mL. Based on that and the imaging findings of abdominal ultrasound (US), Computed Tomography (CT) and Magnetic Resonance (MR) the diagnosis of a recurrent retrorectal pelvic hydatid cyst was made. Three courses of oral albendazole treatment were administered and sixteen weeks later, the patient was admitted for a planned elective operation. At that time, a new CT scan revealed disappearance of the cyst, while the serological tests showed a decrease in the IgG anti-echinococcal antibody titres to 0.71 U/mL. Four different species of the Echinococcus tapeworm can produce infection in humans. E granulosus and E multilocularis are the most common, causing cystic and alveolar echinococcosis respectively, while E vogeli and E oligarthrus, have only rarely been associated with human infection. Although surgical resection remains the treatment of choice for hydatid disease, the present case could suggest that especially in cases of recurrent intraabdominal extrahepatic hydatid cyst, treatment with albendazole may lead to disappearance of the recurrent cyst therefore, should constitute a first line therapeutic option prior to any planned reoperation.

Desaparición de Quiste Hidatídico Intra-abdominal Extrahepático Recurrente Tras la Administración de Albendazol Oral

OP Michail, C Georgiou, PO Michail, E Felekouras, I Karavokyros, G Marinos, A Giannopoulos, J Griniatsos

RESUMEN

Un paciente varón de 44 años con una historia médica pasada de una escisión quirúrgica completa de un quiste hidatídico pelviano dos años antes, se presentó con dolores constantes del piso pelviano y un título de anticuerpos IgG anti-equinocóccicos en plasma, de 14.27 U/mL. Sobre esa base y a partir de los hallazgos de las imágenes del ultrasonido abdominal (US), la tomografía computarizada (TC) y la resonancia magnética (IRM), se llegó al diagnóstico de un quiste hidatídico pelviano retrorectal recurrente. Se administraron tres ciclos de tratamiento con albendazol oral, y dieciséis semanas después, el paciente fue ingresado para una operación electiva planificada. En ese momento, un nuevo escaneo mediante TC reveló que el quiste había desaparecido, en tanto que las pruebas de serología mostraron que el título de anticuerpos IgG anti-equinocóccicos en plasma disminuyó a 0.71 U/mL. Cuatro especies diferentes de tenia equinococo pueden producir infección en los humanos. E granulosus y E multilocularis *son las más comunes, mientras que* E. vogeli *y* E. oligarthrus, *sólo raras veces han sido asociadas con la infección humana. Aunque la resección quirúrgica sigue siendo el tratamiento preferencial para la enfermedad hidatídica, el caso presentado podría sugerir que sobre todo en los casos de quiste hidatídico intra-abdominal extrahepático recurrente, el tratamiento con*

From: First Department of Surgery, Medical School, University of Athens, "Laikon" Hospital, Athens, Greece.

Correspondence: Dr J Griniatsos, 43 Tenedou Street, GR 113-61, Athens, GREECE, Fax: 0030-210 745-6855, e-mail: johngrin@hotmail.com.

albendazol puede llevar a la desaparición del quiste recurrente, y por consiguiente, debe constituir una opción terapéutica de primera línea antes de planificar algún tipo de re-operación.

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INTRODUCTION

Hydatid disease is a worldwide zoonosis caused by the larval stage of the Echinococcus tapeworm which belongs to the family Taeniidae. Four species of Echinococcus produce infection in humans: *E granulosus* and *E multilocularis* are the most common, causing cystic echinococcosis and alveolar echinococcosis, respectively, while *E vogeli* and *E oligarthrus*, cause polycystic echinococcosis but have only rarely been associated with human infection (1).

E granulosus is commonly seen in the grazing regions of the world, including the Mediterranean region, causing the most frequently encountered type of hydatid disease in humans (2). The prevalence of infection varies widely among different regions, ranging between 1 and 220 cases per 100 000 population in certain regions (3). Hydatid disease constitute a significant public health problem in the Middle East, South and Central America, China, former Soviet Union and some sub – Saharan countries (4).

The life cycle of Echinococcus involves a definitive and an intermediate host. Humans are accidental hosts and do not play a role in the biological cycle (1).

E granulosus is a small (3–5 mm long) tapeworm that resides in the jejunum of the dog (definitive host) and other canines and produces eggs that are trapped in the stool. The definitive host may be infected with thousands of worms. The life cycle begins when worms in the intestine of the dog produce eggs, which are then expelled in the faeces of the definitive host and are released to the environment. They are infective to susceptible intermediate hosts and can also incidentally infect humans. Mammals, usually sheep but occasionally humans, ingest the eggs. After ingestion of the eggs by the intermediate host, digestive enzymes liberate an embryo in the duodenum, that passes through the intestinal mucosa to the portal circulation and migrate to visceral organs. As the liver filters out most of the larvae, most echinococcal cysts occur in the liver. Later on, a fluid-filled cyst develops, that differentiates into multiple layers to become a hydatid cyst. In fact, many larvae are killed by the host's defence mechanisms and thus fail to form cysts, but some larvae pass through the liver to lodge in the lung and other organs, where they form cysts. The life cycle is completed when dogs eat mammalian tissues that contain these echinococcal cysts (2, 5, 6). Because two mammalian species are required for completion of the life cycle, direct transmission of echinococcosis from human to human does not occur.

Peritoneal echinococcosis is almost always secondary to hepatic disease, although some unusual cases of primary peritoneal involvement have been described (7–10).

The overall frequency of intra-abdominal extrahepatic hydatid disease ranges between 6.9% (11) and 19% (9).

Recurrent hydatid cyst is defined as a new growth of an echinococcal cyst after complete surgical removal of the primary one (12) and irrespective to the surgical technique; its incidence has been reported as high as 23.3% (12, 13).

CASE REPORT

A 44-year old male patient presented to hospital complaining of constant pelvic floor pain and dysuric symptoms over the last two months. His past medical history revealed an exploratory laparotomy through a lower midline incision and excision of a 10 × 12 × 8 cm presacral cyst, in a district hospital, two years earlier. At that time, the histology report disclosed a completely excised hydatid cyst. His postoperative recovery was uneventful and he was discharged on the 6th postoperative day. He received six courses of oral albendazole for a total period of approximately eight months.

On admission, the clinical examination was unremarkable, haematological and biochemical blood tests were reported as within normal limits, while the plasma IgG anti-echinococcal antibody titre was 14.27 U/mL (for a negative result, titre < 0.9 U/mL is required) (14).

Abdominal computed tomography (CT) scan revealed a 4 x 3. 5 x 3 cm cyst at the level of 2nd–3rd sacral vertebra causing partial obstruction of the left ureter with proximal ureter distention (Fig. 1a). Abdominal magnetic resonance

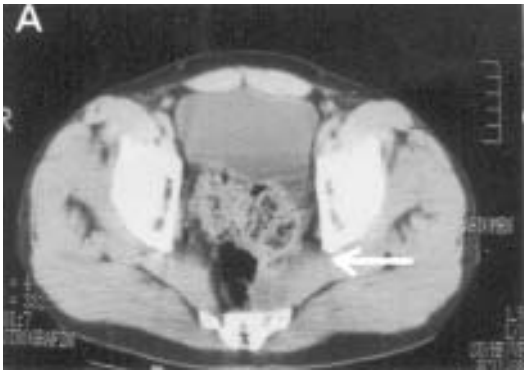

Fig. 1a: Appearance of the cystic lesion at the level of 2nd and 3rd sacral vertebra before the treatment (CT view).

imaging (MRI) scan disclosed a cyst at the same level (Fig. 1b). Finally, an endorectal ultrasound disclosed a 4 x 3 5 x 3.2 cm retrorectal pelvic cyst with smooth border, 10 cm from the dentate line, attached to the sacral bone causing external pressure to the left border of the rectum, without evidence of lymph node enlargement (Fig. 1c).

Flexible cystoscopy did not identify any anatomical abnormality, while urodynamic test revealed significant resi-

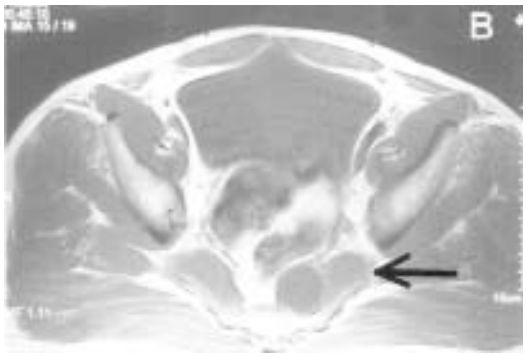

Fig. 1b: The same cystic lesion on the MRI scan of the lower abdomen and pelvis.

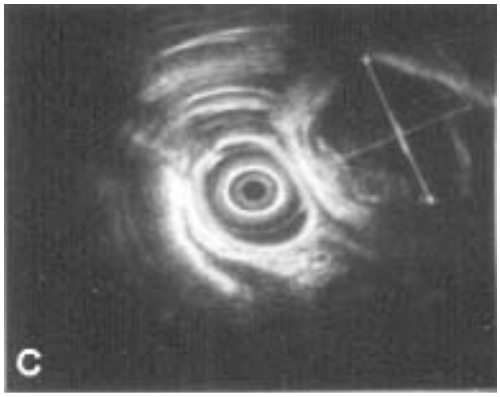

Fig. 1c: Appearance of the cyst in the endorectal ultrasound scan.

dual urine volume, probably related to either sacral nerve damage secondary to the previous radical operation or external pressure on the bladder secondary to the cyst presence.

Based on the above results, a diagnosis of local recurrence of the hydatid cyst was established.

The pelvic floor pain was successfully controlled by the occasional oral use of non-steroidal anti-inflammatory drug (NSAID). Meanwhile, three courses of oral administration of 800 mg of albendazole twice daily (each course consisted of four weeks albendazole administration followed by two weeks discontinuation of the regimen) prior to a planned re-laparotomy for cyst excision at the end of the courses was advocated.

Sixteen weeks later, the patient was re-admitted in order to undergo an elective operation. A new abdominal CT scan was performed at that time and both the previous pelvic cyst as well as the ureter distention had disappeared (Fig. 1d), while the plasma titre of the IgG anti-echinococcal antibodies had decreased to 0.71 U/mL.

DISCUSSION

Since recurrence of an echinococcal cyst mainly follows a silent clinical course, unless its growth produces pressure symptoms or there are complications (13), its diagnosis is

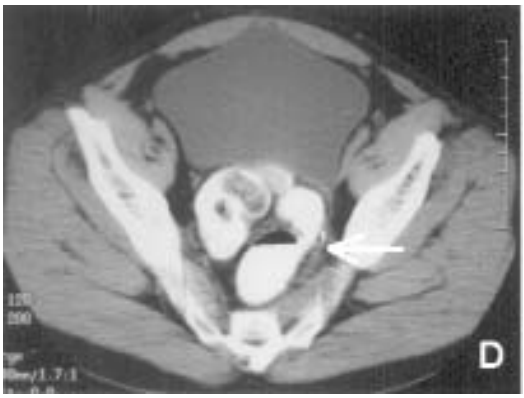

Fig. 1d: CT scan of lower abdomen and pelvis revealing the disappearance of the cystic lesion following three courses of oral administration of albendazole.

commonly based on the results of specific serological tests and the imaging findings (15).

The detection of IgG circulating anti-echinococcal antibodies in serum by the ELISA method constitutes the most sensitive (94%) and specific (99%) test for the majority of cyst locations (16). Test titre increases in the majority of patients for the first three months after surgery (probably as a result of antigen liberation during cyst manipulation) and decreases from the 3rd month after surgery onwards, in patients without relapse (17).

Ultrasonography (US) should be the first imaging choice in abdominal hydatid cysts with sensitivity rates between 93 and 97% (11). Abdominal (CT) scan should be performed in cases of uncommon location of the disease and before any surgical intervention (15). Several reports (18, 19) suggest that CT has a higher overall diagnostic accuracy compared to ultrasound, with sensitivity rates between 95% and 100%. Magnetic resonance imaging is usually not required and in most instances is not cost effective compared to the CT (20). The usefulness of the percutaneous aspiration for diagnostic and therapeutic purposes in cases of extrahepatic location of hydatid cysts cannot be established on the results of small series (21, 22).

The treatment of both primary and recurrent hydatid disease is mainly surgical, as antihelminthic chemotherapy alone has failed in many cases (23). However, WHO recommendations state that medical therapy should be used for: patients with inoperable disease, patients with multiple cysts in two or more organs, patients with peritoneal cysts, patients following incomplete surgery or relapse and for prevention of secondary spread of echinococcal infection following spontaneous rupture or aspiration of cysts (24).

Benzimidazoles (mebendazole and albendazole) are specific chemotherapeutic agents, inhibiting the assembly of tubulin into microtubules, thus impairing glucose absorption through the wall of the hydatid parasite. This causes glycogen depletion and degeneration of the endoplasmic reti-

culum and mitochondria of the germinal layer of the metacestode and results in an increase in lysosomes and subsequent cellular death (25). Several reports (26, 27) suggest that albendazole is more effective than mebendazole, probably because of its more favourable pharmacokinetic profile which leads to higher serum and cyst fluid concentrations. The adverse effects of anti-helminthic therapy are reversible, because they are rarely serious and always subside after discontinuation of the treatment (12).

The index case was operated on two years previously for a pelvic cyst which was histologically confirmed to be a hydatid one. The differential diagnosis of the newly developed cystic lesion in the same area should include either abscess formation (but the presenting symptoms and the CT findings excluded that diagnosis) or chronic haematoma formation (but the elapse period was estimated as too long) as well as development of a malignant soft tissue tumour with central necrosis (but the lack of thick and irregular borders in the CT was incompatible with this). Lymphocoele formation secondary to previous operation could be a possibility but the simultaneous elevation of serum IgG circulating anti-echinococcal antibodies, more than three months since the operation, was highly suggestive of recurrence of the hydatid disease.

In conclusion, although surgical resection remains the treatment of choice for hydatid disease, the present case could suggest that especially in cases of recurrent intra-abdominal extrahepatic hydatid cyst without concomitant hepatic cyst(s) presence, treatment with albendazole may lead to disappearance of the recurrent cyst and therefore, should constitute a first line therapeutic option prior to any planned re-operation.

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