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Case Report of Combined Echinococcosis: Diagnostic and Treatment Peculiarities

Клинический случай сочетанного эхинококкоза: особенности диагностики и лечения

Abstract

The article presents incidence of echinococcosis in humans. There is also modern classification according localization of lesion, diagnostic and treatment peculiarities in combined cases. We presented a clinical case of combined hydatid disease of liver and lung, that was challenge because it made difficulties in surgical treatment, as it was associated with considerable mortality and recurrence rates. Although, there was not compliance of the patient for the surgery as should be performed two kinds of surgery – liver and lung. Was decided to perform only anthelmintics treatment by three cycles of albendazole. It was successful and without any relapses that was shown on the control US and MRI three month after the treatment.
Keywords: Echinococcosis, case report, hydatid disease of liver, hydatid disease of lung.

Резюме

В статье представлена частота заболеваемости эхинококкозом, современная классификация по локализации поражения, а также диагностические и лечебные особенности при комбинированных случаях. Мы представили клинический случай комбинированного эхинококкоза печени и легкого, который был проблематичным, поскольку затруднял хирургическое лечение, что связано со значительной смертностью и частотой рецидивов. Не было приверженности пациента к хирургическому лечению, поскольку необходимо выполнять два вида операции – на печени и легком. Было решено проводить только антигельминтное лечение – три цикла альбендазола, которые проведены успешно, без рецидивов, что было показано при контрольных УЗИ и МРТ-исследованиях через три месяца после лечения.
Ключевые слова: эхинококкоз, история болезни, эхинококкоз печени, эхинококкоз легкого.

■ BACKGROUND

Echinococcosis is biohelminthiasis caused by larval stage of the tapeworm E. granulosus that is characterized by chronic course with solitary or multiple cystic formations in liver, sometimes, in lungs and other organs. In the world medical literature and ICD-10 in the term "echinococcosis" (B.67) two diseases are combined that are caused by different species of helminths of the genus Echinococcus: hydatid echinococcosis or simply echinococcosis

К содержанию

(a causative agent – the larval stages of Echinococcus multilocularis, Echinococcus vogeli and Echinococcus oligarthrus). In our country, these invasions have traditional names "echinococcosis" and "alveococcosis", therefore, we will use these terms hereinafter.

Echinococcosis is dangerous zoonotic parasitic disease worldwide, mainly distributing in Mediterranean regions, Russia, central Asia, China, Australia, South America, and north and east Africa [1, 2]. Annual incidence rates of diagnosed human cases per 100,000 inhabitants vary widely in each country, from less than 1 case per 100,000 to high levels. Cystic echinococcosis is rarely fatal. Occasionally, deaths occur because of anaphylactic shock or cardiac tamponade in heart echinococcosis [2]. The relevance of echinococcosis in Ukraine is due to chronic course of disease, late diagnosis, damage to various organs, increasing number of patients with combined lesions (liver and lungs), and with atypical localizations (kidney, spleen, brain, etc.), high risk of postoperative complications and relapses [3–5].

Cystic echinococcosis is one of the few parasitic infections in which the basis for laboratory diagnosis is primarily serology [6]. Indirect hemagglutination test (IHT) and enzyme-linked immunosorbent assay (ELISA) are the most widely used methods for detection of anti-Echinococcus antibodies (immunoglobulin G [IgG]). But approximately 10% of patients with hepatic cysts and 40% with pulmonary cysts do not produce detectable serum IgG antibodies and exhibit false-negative results.

That's why instrumental methods (Ultrasonography (US), Computer Tomography (CT) scanning, Magnetic resonance imaging (MRI) are basic for detection of echinococcosis and in combination with serological tests let diagnose the disease in most cases. Radiographic examination is useful for cysts in the lungs, bone, and muscle and for detecting calcified cysts.

Ultrasonography is the procedure of choice when making the diagnosis of asymptomatic cystic echinococcosis because it is safe, noninvasive, and relatively inexpensive and is useful in longitudinal studies, such as monitoring the response of cysts to treatment and recording cyst growth rate [7]. Ultrasonographic appearance of echinococcal cysts is seen in the image below (Fig. 1). If ultrasonography cannot be performed owing to cyst location or patient-specific reasons, MRI with heavily T2-weighted series is preferable to CT [9].

Speaking about treatment, surgery was the only treatment available before the introduction of anthelmintic drugs. It is considered the first choice of treatment for echinococcosis but is associated with considerable mortality (up to 2% in some series, increasing with second and further operations), morbidity [10] and recurrence rates (2–25%). Given the more frequent detection of early and asymptomatic E. granulosus liver lesions, a widened indication for chemotherapy exists. Several procedures have been described for the treatment of hepatic echinococcal cysts, ranging from simple puncture to liver resection and transplantation, although the most commonly used technique is total or partial cystopericystectomy. Surgery for pulmonary cysts includes extrusion of cysts using Barrett technique (intact endocystectomy without preliminary aspiration), pericystectomy, and lobectomy [11].

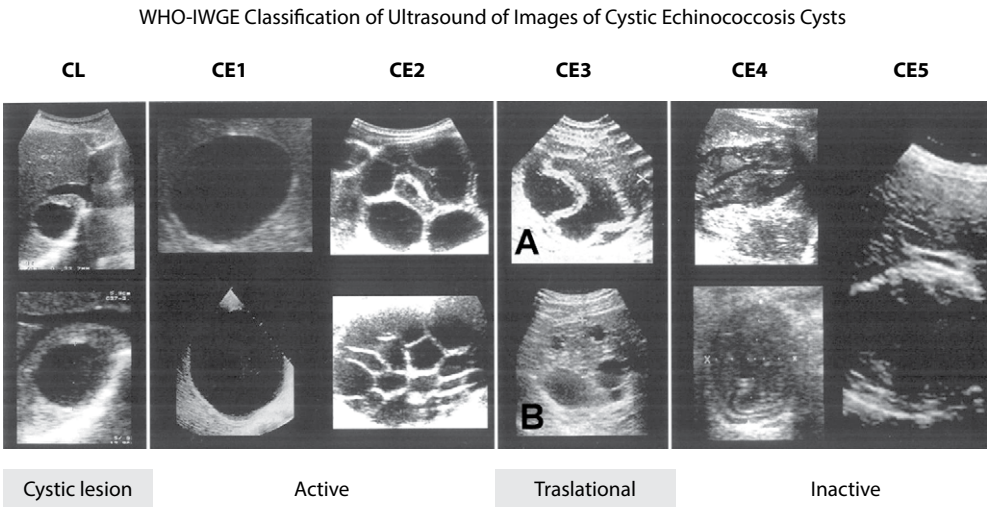

Fig. 1. WHO Informal Working Group on Echinococcosis standardized ultrasound classification of echinococcal cysts. Image of World Health Organization (WHO) [8]

The drug of choice against echinococcosis is albendazole because its degree of systemic absorption and penetration into hydatid cysts (is superior to that of mebendazole, for example). Albendazole in combination with percutaneous aspiration can lead to a reduction in cyst size [12]. When surgery cannot be avoided, presurgical use of albendazole in echinococcus infestations reduced risk of recurrence and facilitated surgery by reducing intracystic pressure.

However, the aim of this case report is to present a rare case of combined echinococcosis of liver and lung that treated successfully without surgery. A 59-year-old man presented with upper abdominal discomfort and pain, poor appetite, chronic cough and dyspnea in physical exercise. There was a history of contact with cattle and dogs. Systemic history was unremarkable.

The physical examination was normal except hepatomegaly. Laboratory tests were normal. US examination was impaired because of bowel gas and obesity but it was revealed simple oval unilocular cyst with anechoic content and a visible double cystic wall and floating membranes, i.e. the endocyst detached from the cyst outer wall (pericyst); its size was 5,6*5,3 cm (CE 3a according WHO classification) in right lobe of liver and sings of steatosis of the liver. (Fig. 2).

A radiograph of the chest revealed a lesion of the upper lobe of the right lung. For confirming the diagnosis serologic test ELISA was performed and detected high level of IgG to E. granulosus. MRI of chest and abdomen was also indicated for exclusion disseminated disease and complications. There was detected round not permeable formation which has a thin capsule 2×3 cm in size in the upper lobe of the right lung between the 3rd and 1st segments. Its vascularization was moderate. Other sites of localization were excluded.

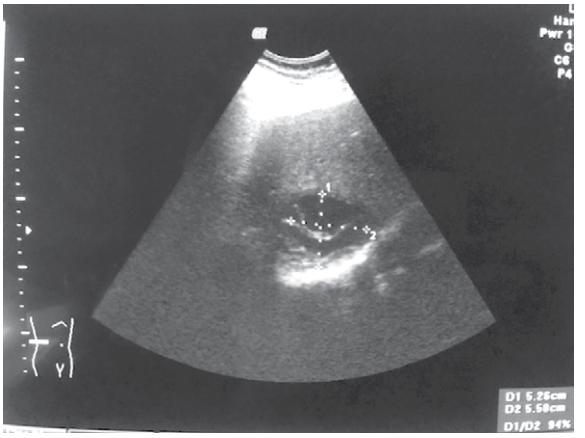

Fig. 2. Simple oval unilocular cyst with anechoic content and a visible double cystic wall and floating membranes

So the diagnosis was combined echinococcosis of liver and lung. He got the treatment with albendazole administered PO with meals in a dose of 400 mg twice daily for 28 days total of 3 treatment cycles after 14 days without treatment between each cycle. Speaking about surgical treatment, there was no compliance of the patient.

Control US of abdomen was performed after 3 cycles of albendazole and the cyst became uniformly echogenic, but it was hypoechogenic (CE4 according WHO classification) with the same sizes (Fig. 3). That exhibited involution of cyst content and is nearly always found to be non-viable. Though the patient was recommended surgeon consultation for surgical treatment of the cyst as it was more than 5 cm in diameter. Unfortunately the patient refused surgery.

There was not recurrence of hydatid cyst at three months follow up of the patient.

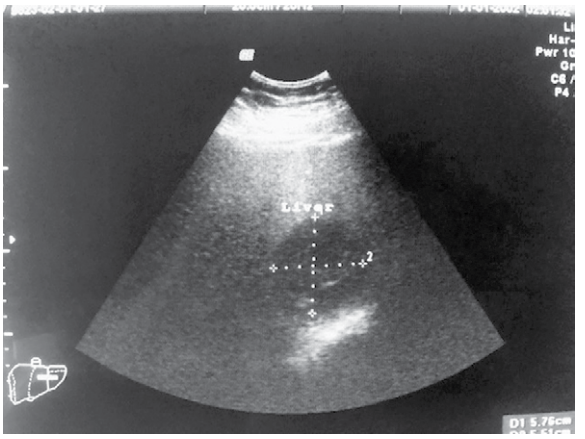

Fig. 3. Uniformly echogenic cyst of the patient after 3 cycles of albendazole treatment

■ CONCLUSION

Echinococcal cysts have to be differentiated from other conditions, such as non-parasitic cysts, single or multiple hemangiomas, pyogenic or amoebic liver abscesses, hematoma, and neoplasia with hemorrhage and necrosis. Determining whether a cystic lesion is echinococcal depends on the presence of a double wall and is obvious when membrane detachment is present. And positive serology test also helped us in correct diagnosis.

The challenge in this case laid in combined localization of the cyst in liver and lungs. That's why it makes difficulties in surgical treatment, because it was associated with considerable mortality, morbidity, and recurrence rates. Although, there was not compliance of the patient for the surgery as should be performed two kinds of surgery – liver and lung. And we decided to perform only anthelmintics treatment by three cycles of albendazole. And it was successful and without any relapses that was shown on the control US and MRI three month after the treatment.

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