

## Case reports

# Cystic echinococcosis in a child infected with HIV

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**ABSTRACT.** Echinococcosis is a parasitic disease caused by the tapeworm *Echinococcus granulosus*. *Echinococcus* infection is possible at any age, including childhood. Most of the cases are recognized accidentally. HIV infection in children is rarely diagnosed in Poland. A currently 16-year-old girl was diagnosed with HIV vertical infection at the age of 13. Antiretroviral therapy was started after 6 months of observation. Routine ultrasound examination of her abdomen revealed a cystic lesion in the liver. The IgG ELISA test for *E. granulosus* infection was negative. However, she was treated with albendazole due to clinical suspicion of echinococcosis. After anti-parasitic treatment, an abdominal ultrasonography (US) and computed tomography (CT) scans were performed and revealed progression of the lesion (one year of observation). As an additional imaging study, biliary tract scintigraphy was done. Localization of the cyst allowed its surgical removal. The surgery was performed under pharmacological protection with albendazole. Histopathology examination confirmed the diagnosis of echinococcosis. Currently, the clinical condition of the patient is good, antiretroviral treatment is effective and repeated abdominal ultrasound is unremarkable. *E. granulosus* infection in children is rare and may be accompanied by other diseases and infections. Diagnosis is difficult and it is often based on the clinical picture without serological confirmation. Surgical treatment should be supplemented with pharmacological treatment.

**Key words:** children, HIV, *E. granulosus*

## Introduction

Echinococcosis is a parasitic disease caused by the tapeworm *Echinococcus granulosus*. The infection occurs during the accidental ingestion of eggs with contaminated soil from hands or food products. Larva enters the human internal organs and, while developing, it causes clinical symptoms similar to cyst or tumor. The cyst is usually located in organs such as liver and lungs but also in spleen and kidneys [1–3].

Human cystic echinococcosis (CE) remains highly endemic in pastoral communities in such regions as East-Central Europe, Russia, the Mediterranean basin, the Near and Middle East, Central Asia, China, East Africa and South America [2,3].

Annual incidence rate in Poland is low (40 cases in 2007, 28 in 2008). Provinces with the highest incidences are warmińsko-mazurskie, podlaskie and

lubelskie. Additional risk factors are of occupational nature (foresters, farmers) [5].

Infection of *Echinococcus* is possible at any age, including childhood (no epidemiological data are available for this age group). Most of the cases are detected accidentally during routine ultrasound (US), more rarely – as a consequence of evaluation of patient's symptoms and signs [3,4]. WHO-IGWE (Informal Working Group on *Echinococcus*) classification is used to describe cystic lesions found on US. It divides cysts into following groups: CL – cystic lesion, which is undifferentiated; CE 2, CE 3 – early stages; CE 4, CE 5 – late stages [4].

HIV infection in children is rarely diagnosed in Poland. It may be accompanied by other diseases, not related to retroviral infection. About 90% of HIV infection cases in children is due to mother-to-child transmission [6–8]. 50% of them are diagnosed late (above the first year of life) and 25% at the stage of clinically overt AIDS.

The aim of this study is to present a case of 16-year-old HIV-infected girl with an accidentally diagnosed CE.

### Case presentation

A 16-year-old girl was born by spontaneous vaginal delivery at 37 weeks of gestation, from a twin pregnancy, estimated at 9–10 points on the APGAR scale. She was formula fed. The neonatal period was uneventful. Her twin brother died in the first year of life from unknown causes (probably due to AIDS). The girl lives in the lubelskie province. At the age of 13 she was found to be HIV-infected (after her mother had been diagnosed with AIDS) at the stage of mild clinical symptoms and without immunodeficiency. After 6 months of observation, combined antiretroviral therapy (cART) was started. Atripla (tenofovir+efavirenz+emtricitabine) was introduced. The treatment was tolerated well. The routine ultrasound of her abdomen revealed an uncharacteristic liver cyst, which corresponded with stage CL on WHO classification (January 2007). Subsequent studies (July 2008) showed thickening of the cyst wall (stage CE3). The IgG ELISA test for *E. granulosus* was negative (April 2008). Basing on the clinical picture and imaging studies she was treated with albendazole (600mg/daily) for 4 weeks. After the anti-parasitic therapy abdominal CT scan was performed and revealed further progression of the lesion – it was hypodense with calcifications located circumferentially, which was consistent with the *E. granulosus* cyst – CE 4/5 (July 2008). Subsequent US showed no improvement (April 2009, July 2009). As an additional imaging test, biliary tract scintigraphy was performed, which showed no communication between the liver cyst and bile ducts (October 2009). Localization of the cyst allowed its surgical removal. The procedure was performed under pharmacological protection with albendazole (October 2009). At that time the girl was without immunodeficiency, with an undetectable HIV viral load. Perioperative cART was continued with soluble formulations of the drugs. Treatment with albendazole was continued for 2 weeks after surgery. Postoperative histopathological examination of the cyst confirmed *E. granulosus* etiology. The patient did not report any complaints. At present, the girl is in good clinical condition, antiretroviral treatment is effective and no lesions are found on repeated abdominal US.

### Discussion

Diagnosis of echinococcosis is difficult and based on clinical findings, imaging studies and serology. Clinical manifestation depends primarily on localization and size of hepatic lesion and may include hepatomegaly, obstructive jaundice or cholangitis [3,9,10]. The most common imaging study is an abdominal US examination, which can show a specific cystic pattern of the lesion. It is non-invasive and repeatable, the results are real-time and it does not require special preparing of the patient [3,9,10]. For cystic echinococcosis (CE), choosing the treatment (percutaneous endoscopy, surgery, drug treatment or observation – „watch and wait”) is stage-specific and based on imaging studies. It is recommended to use an imaging-based WHO-IGWE classification. In our case in the first US, the image was nonspecific and gave a suspicion of parasitic infection – CL stage [4]. Further imaging examinations (US and CT) confirmed the diagnosis – C3 and C4/5. Serological tests are important in the differential diagnosis but a negative result does not exclude the infection. The specific IgG ELISA tests are of 26–60% sensitivity. These tests are not specific for *E. granulosus* so it might give false-positive results in patients with other parasitic infections, such as alveolar echinococcosis, cysticercosis or schistosomiasis [2,11,12].

The most sensitive (96.5 %) is specific IgG ELISA AgB (antigen B-rich fraction) which is not a standard test in Poland [11,12]. After the first US, as the echinococcosis etiology was suspected, IgG ELISA test was performed in our patient but the result was negative.

It has to be mentioned that children infected with HIV have impaired antibody response which can be an additional cause of negative results of serological tests [13,14]. The final diagnosis is intra- and postoperative histopathological examination. Postoperative examination confirmed the diagnosis of echinococcosis in our patient.

Surgical treatment is a treatment of choice because it enables a radical excision of the cyst. Conservative procedures, relatively simple, are still accepted, for example PAIR (puncture, aspiration, injection, reaspiration) or laparoscopic surgery which has a higher rate of complications. These procedures are not performed in children. Surgical therapy should be supplemented with pharmacological treatment [9,13,15,16]. Sometimes the cyst cannot be surgically removed because of the

extension of occupied area, the lack of surgical access or other tissues proximity and the danger of their damage [15–17]. Accidental rupture of the cyst during the operative removal or due to mechanical trauma (by larger size of the lesion) can lead to secondary echinococcosis and/or anaphylactic shock. Therefore, the surgical treatment is carried out under pharmacological cover [9,13,16,17]. The localization of the cyst in our patient enabled its total resection. The peri- and postoperative period was uneventful.

Pharmacological treatment does not always bring expected and desired effect. The first benzimidazole, which was used in the treatment of echinococcosis, was mebendazol. However, it was associated with treatment failure in some cases because of its poor absorption. The drug which is recommended according to the literature is albendazole. In some cases, such as nonresective cysts, it is recommended to use a combination of mebendazol/albendazol with praziquantel [15,16,18]. In our case, albendazol was used for 4 weeks after the second US (April 2007). The second course of treatment was started two weeks prior to the surgery and continued for 2 weeks thereafter. In control US after the operation, no cysts or other lesions were found.

When HIV is diagnosed, the patient is screened for opportunistic infections. Clinical manifestation of parasitic infestation may be similar to them. Hence, it is important to take such conditions into consideration in differential diagnosis [13].

## Conclusions

*E. granulosus* infection in children is rare and may be accompanied by other diseases and infections. The diagnosis and treatment of echinococcosis is difficult. The effectiveness of medical intervention is individually variable.

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