

Case Report

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Long-Term Drug Treatment of a Giant Liver Hydatid Cyst in a Child

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ABSTRACT

Background: Cystic echinococcosis is a chronic parasitosis caused by the larvae of the tapeworm *Echinococcus granulosus*. Humans act as intermediate hosts and the larvae can affect any organ in the form of cysts. They are most often found incidentally by diagnostic imaging, where they have become large in size, with treatment being primarily invasive. Conservative treatment with albendazole is one of the appropriate treatment options, but it features a low success rate in cysts over 5 cm.

Case Presentation: We describe a case of an 16-year-old girl with an active 12 cm cyst in the liver. Two consecutive courses with albendazole, with duration 3 months each, were conducted. The cyst reached a safe inactive stage and no relapses have been observed for 1 year since the last administration of the medication.

Conclusions: Our experience shows that with large cysts albendazole courses may be extended including in childhood. Ultrasonography has clearly demonstrated its ability to follow-up patient in a cost effective manner and avoiding ionizing radiations.

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Introduction

Cystic echinococcosis (CE) is a severe parasitic zoonosis caused by the larvae of the tapeworm *Echinococcus granulosus*. It is spread almost globally - endemic areas in Europe are the Balkans and the Mediterranean. Definitive hosts are mainly dogs and intermediate hosts are ruminants (sheep). Because of their proximity to these animals, humans may be accidental intermediate hosts. The larval form called hydatid cyst can affect any tissue or organ, but the liver is most commonly attacked. The course of the disease is slow and asymptomatic, and the disease is most often diagnosed incidentally at an advanced stage. The pediatric population accounts for 10–20% of all cases [1, 2]. Depending on the duration of the disease, liver cysts have different sonographic characteristics and dimensions (Table 1), based on which, one of four approaches is recommended - medication treatment, open surgery, cyst puncture or “watch and wait” for cysts in a transitional stage [3].

Table 1: Different Sonographic Characteristics of Liver Cysts, World Health Organization-Informal Working Group Classification on *Echinococcus* (Who-Iwge).

Stage Echographic image According to Who-Iwge Classification
CL Anechogenic uniloculated cyst, with no echoes or internal sepsis
CE1 Anechogenic cyst, with fine echoes inside, representing the hydatid sand - active cyst
CE2 Cyst with multiple septums at the interior, or “honeycomb” aspect, with a uniloculated primary cyst - active cyst
CE3 Uniloculated cyst with decolated membrane (“waterlily sign”) (CE3a) or daughter vesicles type hypo/hyperechogene images (CE3b) - cyst in transition phase
CE4 Cyst with mixed content, hypo/hyperechogenic, without daughter vesicles - degenerative phase
CE5 Cyst with partial or totally calcified wall - inactive cyst

Case presentation

We report a case of an 16-year-old girl with an active 12 cm cyst in the liver. Diagnosis was obtained by ultrasonography (US) (Samsung Medison Convex Array Ultrasound Transducer 3–7 MHz.) and serologically with enzyme-linked immunosorbent assay [ELISA] immunoglobulin G [IgG]; BulBio-National Centre of Infectious and Parasitic Diseases, Sofia, Bulgaria). The present study was approved by the ethical committee of University Hospital “Prof. Iv. Kirov” and Medical University of Sofia (number 12/2018). A 14-year-old girl living in a rural area in southern Bulgaria visited her family doctor because of complaints of fatigue and heaviness in the right hypochondrium. On examination, the doctor found mild tenderness upon palpation in the liver area and elevated eosinophil level in the peripheral blood (11.4%). Due to eosinophilia, the patient was referred to our clinic with suspected parasitosis. Abdominal sonography showed an active echinococcal cyst in the right lobe of the liver, 12 cm in diameter. By the WHO classification, the cyst was in stage CE3a (with daughter cysts) [4]. Serological testing of the patient was positive for echinococcosis. Due to the patient’s parents’ refusal of surgery, long-term albendazole treatment (15 mg/kg for 3 months, without interruption) was initiated. The follow-up sonography, after the 3-month treatment course, showed that the cyst had reached a safer stage CE3b - transitional cyst. The medication treatment was discontinued for one month, and then re-initiated, with albendazole at the same dose for 3 months. A month of rest and a new three-month course with the medication followed. Upon completion of the third course of medication treatment, the sonographic examination showed that the cyst had reached CE4 stage - inactive cyst (Figure. 1). Liver transaminase levels were observed throughout the course of medication treatment, and they did not rise above normal. Presently, the patient has been having an inactive cyst for 1 year, and sonographic monitoring is performed using the “watch and wait” approach. Serological testing is still positive.

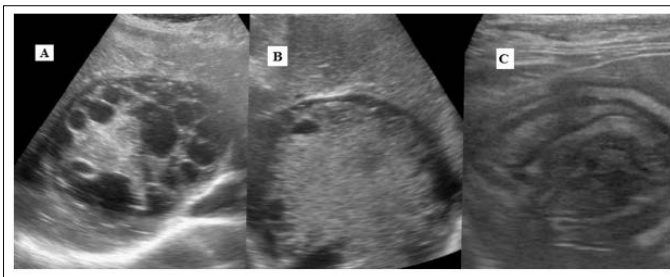


Figure 1: A. Before treatment, stage C3a, B. After the 1st course of treatment, stage C3b, C. After the 3rd course of treatment, stage CE4.

Discussion

CE is most often found incidentally during prophylactic examination or due to non-specific complaints. At the time of diagnosis, the cyst is most often relatively large, active, in some cases, two or more cysts are found - multiple echinococcosis. Treatment is very important to avoid complications [4]. Conservative treatment with albendazole is one of the appropriate treatment options, but it features a low success rate in cysts over 5 cm in diameter, probably due to the more difficult penetration of the active substance into the cyst [5, 6]. In such cases, surgery or puncture method are preferred. Despite their good results, these two invasive methods often have contraindications or patients refuse to have them administered because they would like to try medication. The WHO informal echinococcosis group recommends continuous medication treatment for 1 month [7, 8]. Some authors accept that

albendazole therapy for <3 months is associated with suboptimal outcomes and some authorities propose even longer therapies (>6 months), although this approach requires further validation [9, 10]. In the case described, we administered longer courses because of the size of the cyst and the risk of rupture (active and superficial). Our experience shows that in some cases albendazole courses may be extended if the patient has good tolerability to the medication. In inoperable cases, conservative treatment may also be applied to larger cysts, including in childhood. Adverse reactions have been associated with non-adherence and discontinuation of therapy and have been found to affect adherence to visits in conditions requiring prolonged therapy [11]. Despite CT (Computed tomography) or MR (Magnetic resonance) are often used, in this case US has clearly demonstrated its ability to follow-up patient in a cost effective manner and avoiding ionizing radiations [5].

Conclusion

CE remains a serious problem in some endemic areas of Europe, such as the Balkans and the Mediterranean. Along with extensive sheep breeding and many stray dogs, in some of these areas, veterinary measures for control of sheep and deparasitization of dogs as a major preventive measure against parasitosis are underestimated.

Authors’ contributions

VV wrote the manuscript; VGV designed Tab. 1 and helped to write the manuscript; VV diagnosed and treated the patient; YMM and VGV helped to epidemiological study and serological testing. VV make a design to Figs. 1; YMM revised manuscript critically for important intellectual content; all authors read and approved the final manuscript.

Ethics Approval and Consent to Participate

Ethics approval was obtained from University Hospital “Prof. Iv. Kirov”. Written informed consent to participate in the study from the patient’s parents was obtained.

Consent for Publication

Written informed consent to publication on the study from the patient’s parents was obtained.

Competing Interests

The authors declare that they have no competing interests.

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