

Case Report: Percutaneous Treatment of Multiple Echinococcal Cysts Presenting as Abdominal Palpable Mass

Chiara Tersigni,¹ Alessandro Semeraro,² Marcello Caremani,³ Elisabetta Venturini,¹ Claudio Defilippi,² Maurizio de Martino,¹ and Luisa Galli^{1*}

¹Department of Health Sciences, University of Florence, Anna Meyer Children's University Hospital, Florence, Italy;

²Department of Radiology, Anna Meyer Children's University Hospital, Florence, Italy; ³Section of Infectious Diseases, San Donato Hospital, Arezzo, Italy

Abstract. We report the case of an adolescent Moroccan girl with abdominal pain and palpable mass in the upper right side of the abdomen. In the emergency department, an abdominal ultrasound showed hepatomegaly and eight active liver cysts, compatible with cystic echinococcosis. Serology for *Echinococcus granulosus* confirmed the diagnosis. Other sites of localization were excluded. Treatment involved albendazole combined with puncture, aspiration, injection, re-aspiration, performed only for the most medial cysts. Periodical follow-up with abdominal ultrasound and with abdominal magnetic resonance imaging showed a progressive involution of all cysts. The treatment with albendazole was stopped after, overall, 6 months, and monthly ultrasound scan were planned as follow-up. In case of hepatic cysts, *E. granulosus* should be excluded, especially in children from endemic countries. A multidisciplinary approach with pediatric infectious disease specialists, radiologists, and surgeons is fundamental for disease management.

CASE REPORT

We report the case of a 13-year-old girl of Moroccan origin who moved to Italy in June 2017. She was admitted to our institution in July because of the presence of worsening abdominal pain and palpable abdominal mass in the upper right side of the abdomen. Fatigue, loss of appetite, and weight loss were also reported. This mass was present since 1 year and no medical investigations were performed in the country of origin. No fever, abdominal pain, neurological or respiratory symptoms, or contact with dogs, sheep, or other animals were reported in her previous medical history. In the emergency department, an abdominal ultrasound (US) showed hepatomegaly and eight liver cysts, compatible with cystic echinococcosis (CE). The largest cyst measured about 56 mm in diameter with a mass effect on the right kidney and the pancreatic head (Figure 1A). All cysts, except one, appeared active (CE1 according to the World Health Organization classification). The girl was admitted to the pediatric ward and blood tests were performed, which showed no hypereosinophilia and a mild increase of serum immunoglobulin E (IgE). The other blood tests performed, including C-reactive protein (CRP), were normal. Serology for *Echinococcus granulosus* showed total immunoglobulin (Ig) with indirect hemagglutination > 1:8,192, and total Ig with enzyme immunoassays > 1:400.

To exclude other sites of localization, a chest X-ray, a fundus oculi, and a brain computed tomography (CT) scan were performed, all of which produced negative results. To better characterize the hepatic lesions, an abdominal CT scan was performed. Treatment with albendazole (ABZ) at the maximum dosage of 15 mg/kg/daily was started. The girl was evaluated after 2 weeks of medical treatment and puncture, aspiration, injection, re-aspiration (PAIR) of the two most medial cysts was performed using ethanol. About 20 cc of alcohol was used for each of the two cysts and they were punctured with a Chiba needle (18G), and about 60 cc of clear liquid was aspirated

from each cyst. The liquid was sent for laboratory tests and the parasitologic test was positive for protoscolices, confirming the diagnosis. Blood tests showed mild hypereosinophilia (1,728 cell/mm³) and a mild increase of CRP (8.84 mg/dL) after the procedure. No complications related to PAIR were detected. Thereafter, an abdominal US was performed, which showed “detained appearance of the two treated cysts, with detachment of the endocyst and formation of septa. There was no free liquid in the abdomen. The remaining findings were unchanged compared with the previous examination” (Figure 1B). After 10 days, the girl was re-evaluated and blood tests showed the reduction of both hypereosinophilia and CRP (2.89 mg/dL). One month after the procedure, an abdominal magnetic resonance imaging (MRI) scan with the study of the biliary tract was performed and showed “a significant involution of all eight hepatic hydatid cysts with modest volumetric reduction, with detained profiles and exfoliation of the internal parietal portion, with an internal signal mainly hyper-intense in the high-repetition time (TR) sequences and isolated to the liver in T1. At the completion of echo tomography, at least two presented detachment of the endocyst and formation of septa. There were no images compatible with the production of daughter cysts, nor of intra-peritoneal dissemination” (Figure 2A). An abdominal US was also performed (Figure 1C). The girl was periodically followed-up with US which showed a progressive involution of all eight cysts with the medical treatment, and no further alcoholization procedures were required. After about 6 months of treatment, an MRI was repeated, and “of the known eight cysts, only one in the VIII segment shows a mild increase in size of about 2 mm. All cysts appeared inactive with a pseudotumor aspect (CE4 WHO classification)” (Figure 2B). An abdominal US was also performed (Figure 1D). Blood examinations, full blood cell count, serum transaminases, creatinin, and CRP were periodically performed showing normal results. The girl's general conditions dramatically improved with a weight gain of about 9 kg.

Based on MRI scan findings, the treatment with ABZ was stopped after overall 6 months, and monthly US scans were planned as follow-up. No relapses were observed after 3 months of follow-up.

* Address correspondence to Luisa Galli, Department of Health Sciences, University of Florence, Anna Meyer Children's University Hospital, Viale Pieraccini 24, Florence I-50139, Italy. E-mail: luisa.galli@unifi.it

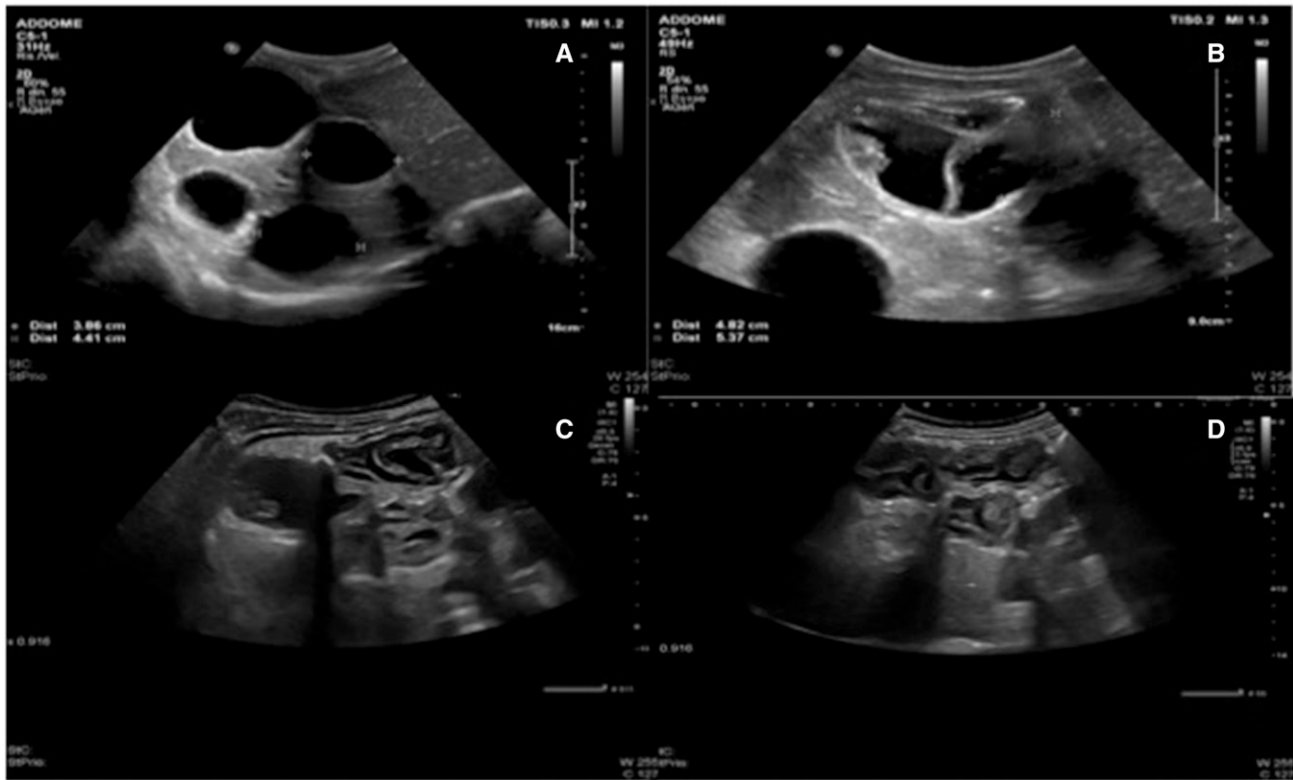


FIGURE 1. Abdominal ultrasound appearance of cysts. (A) Appearance of cysts at the diagnosis; (B) Detained appearance of the two treated cysts. The remaining findings were unchanged; (C) Involution of all eight hepatic hydatid cyst; (D) All cysts appeared inactive with a pseudotumor aspect.

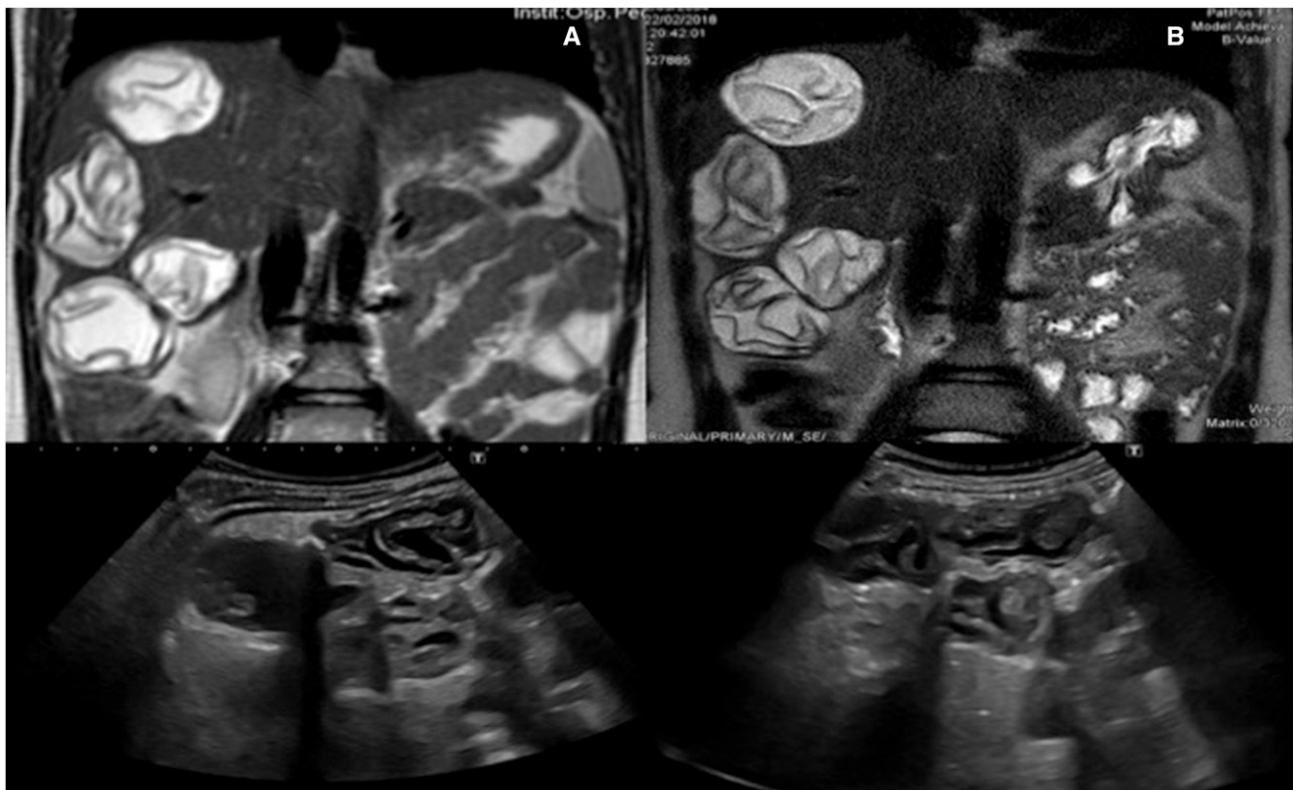


FIGURE 2. Abdominal magnetic resonance imaging and ultrasound of cysts. (A) Magnetic resonance image performed one month after puncture-aspiration-injection-reaspiration procedure. An involution of all eight hepatic hydatid cysts with modest volumetric reduction was observed. At the completion of echotomography, two cysts had thick flaking membranes, giving a thick semisolid aspect. (B) Magnetic resonance image performed six month after puncture-aspiration-injection-reaspiration procedure. All cysts appeared inactive with a pseudotumor aspect.

DISCUSSION

The peculiarity of our case is the presence of eight echinococcal hepatic cysts in a 13-year-old girl. In case of hepatic cysts, the CE etiology should be excluded, especially in children from endemic countries.^{1–3} In case of the presence of several cysts with daughter cysts and high-risk rupture localization, some authors suggest a demolitive surgery approach. However, after accurately considering risks and benefits, and their localization and size, a combined approach with pharmacologic and percutaneous treatment was decided. As matter of fact, according to the literature, no specific indications are available regarding the treatment of multiple active hepatic cysts.^{4–6} In our patient because of the high risk of rupture, the medical treatment alone was not considered safe. In addition, surgery was not indicated because cysts were localized in different hepatic segments. Therefore, PAIR was successfully performed on the two most medial cysts at higher risk of rupture for their superficiality combined with ABZ for 6 months, continued also to prevent secondary CE due to percutaneous treatment. It is pivotal to individualize treatment, especially in complex cases, when a single approach is insufficient. Moreover, a multidisciplinary approach with pediatric infectious disease specialists, radiologists, and surgeons is fundamental for disease management. A regression of all cysts (from CE1 to CE4) was observed, but based on literature, there is a very high risk of relapses. Risk of relapses for cysts with spontaneous involution is about 3%; being 50% for cysts regressed with ABZ.^{6–8} It is important to closely follow-up those patients with monthly abdomen US to detect relapses early.

Received May 31, 2018. Accepted for publication August 22, 2018.

Published online November 12, 2018.

Authors' addresses: Chiara Tersigni, Elisabetta Venturini, and Luisa Galli, Department of Health Sciences, University of Florence, Anna

Meyer Children's University Hospital, Florence, Italy, E-mails: chia88.te@gmail.com, elisabetta.venturini@virgilio.it, and luisa.galli@unifi.it. Alessandro Semeraro and Claudio Defilippi, Department of Radiology, Anna Meyer Children's University Hospital, Florence, Italy, E-mails: alessandro.semeraro@meyer.it and claudio.defilippi@meyer.it. Maurizio de Martino, Department of Health Sciences, University of Florence, Anna Meyer Children's University Hospital, Florence, Italy, E-mail: maurizio.demartino@unifi.it. Marcello Caremani, Section of Infectious Diseases, San Donato Hospital, Arezzo, Italy, E-mail: m.caremani@gmail.com.

REFERENCES

- Center for Disease Control and Prevention, 2012. *Parasites - Echinococcosis - Biology*. Available at: <https://www.cdc.gov/parasites/echinococcosis/biology.html>. Accessed January 14, 2018.
- Brunetti E, Kern P, Vuitton DA; Writing Panel for the WHO-IWGE, 2010. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. *Acta Trop* 114: 1–16.
- Deplazes P et al., 2017. Global distribution of alveolar and cystic echinococcosis. *Adv Parasitol* 95: 315–493.
- Hemphill A, Stadelmann B, Rufener R, Spiliotis M, Boubaker G, Müller J, Müller N, Gorgas D, Gottstein B, 2014. Treatment of echinococcosis: albendazole and mebendazole—what else? *Parasite* 21: 70.
- Moroni S, Moscatelli G, Bournissen FG, González N, Ballering G, Freilij H, Salgueiro F, Altcheh J, 2016. Abdominal cystic echinococcosis treated with albendazole. A pediatric cohort study. *PLoS One* 11: e0160472.
- World Health Organization (WHO), 2001. *PAIR: Puncture, Aspiration, Injection, Re-Aspiration. An Option for the Treatment of Cystic Echinococcosis*. Available at: http://apps.who.int/iris/bitstream/10665/67207/1/WHO_CDS_CSR_APH_2001.6.pdf. Accessed January 14, 2018.
- Rinaldi F, De Silvestri A, Tamarozzi F, Cattaneo F, Lissandrini R, Brunetti E, 2014. Medical treatment versus “watch and wait” in the clinical management of CE3b echinococcal cysts of the liver. *BMC Infect Dis* 14: 492.
- Stojkovic M, Rosenberger KD, Steudle F, Junghanss T, 2016. Watch and wait management of inactive cystic echinococcosis—does the path to inactivity matter—analysis of a prospective patient cohort. *PLoS Negl Trop Dis* 10: e0005243.