

SECONDARY ECHINOCOCOSIS - RUPTURED HEPATIC HYDATID CYST IN CHILDREN

Paun Fuicu¹, Nicolae Babeu², Sebastian Damen², Eugen Sorin Boia¹

Abstract

Hepatic hydatid cyst (HC) is the most frequent location (60%) in children. Besides the direct mechanical action exerted on the affected organ (producing local morphological changes), HC can cause complications, with negative impact on the entire body through its toxic-allergic action [1].

We present a case of a nine years old female child from urban area with hepatic HC, ruptured into the peritoneal cavity with acute peritonitis. Corroborating the clinical data with laboratory, it was suspected the existence of a positive diagnosis of hepatic HC ruptured and discharged into the peritoneal cavity, biliary lithiasis with acute cholecystitis. The biological confirmation of the parasitosis was brought during hospitalization by ELISA test. Surgical exploration showed a large ruptured HC of the left lobe of the liver, with multiple daughter cysts in the peritoneal cavity. Surgical treatment consisted in median laparotomy with hepatic Lagrot procedure, cleaning the ruptured cystic and abdominal cavity with hypertonic saline 20% and its external drainage on silicone tube. The post-operative evolution of the case was without major complications. The hepatic cavity disappearance was found in ultrasound examination in 3 months after the hospital discharge and the biliary lithiasis after 6 months.

The diagnosis of hydatid cyst must be considered especially in endemic regions. We consider that the peritoneal cleaning with hypertonic saline and the early and prolonged prophylaxis with anthelmintics may be salutary in order to avoid secondary echinococcosis.

Keywords: hydatid cyst, hydatid disease, Lagrot procedure, Taenia Echinococcus, child.

Introduction

The hydatid cyst continues to attract the attention of the medical world by the frequency of the cases accidentally discovered in infantile population, or of its complications. It is caused by the larval stage of development of Taenia Echinococcus Granulosus in different human organs (liver 60%, lung 30% and other organs 10%). HC had a torpid evolution for years, or become acute by the rupture of the

cyst followed by mechanical, allergic or septic complications which can endanger the patient's life. [2] [3]

The incidence of the HC in the western part of Romania, is estimated at 3.1 \ 100,000 children, WHO considering our country as an endemic area [4] [5].

In recent years, the urbanization of the disease was observed, probably due to the large number of tramp dogs, or due to the population migration from rural to urban areas [5].

Due to globalization which includes tourism and migration, the condition required an approach also by the countries that showed a low incidence of the disease or where it was considered eradicated (Western Europe, USA) [6] [7].

The rupture of hepatic HC and its evacuation into the peritoneal cavity is rarely mentioned in children literature. HC present some special clinical-evolutive features and, indications for therapeutic methods. [8][9].

Clinical case presentation

We present the case of a nine years old female child from urban area, hospitalized in our service transferred from the infectious disease department.

She presented with *abdominal pain syndrome, acute enterocolitis, acute dehydration syndrome 10% and acute erythematous angina.*

After 48 hours of treatment in infectious disease department, the abdominal pain was not remitted, became colicative and diffuse, with the muscle contraction.

On admission in our service, the patient is conscious, febrile (38°C), with general influenced condition, lose of appetite, nausea and diarrhea. Pulmonary, she presents superficial breaths with tachypnea, tachycardia with pulse 140 beats / min. Mobile abdomen with breathing - spontaneously diffuse - sensitive and at superficial palpation a tumoral hypogastric tumour, with pain and reflex generalized muscle contraction at deep palpation.

Laboratory findings present leukocytosis (14 x 10³) with neutrophilia (69.4%) and eosinophilia (15.6%).

The increased inflammatory samples, ESR 55 mm / 1 h, fibrinogen 5.60 g / l, C reactive protein (PCR) 57.26 mg / l, procalcitonin 0.07 ng / ml, and glucose 7.66 mmol / l.

¹Department of Pediatric Surgery, "Victor Babeş" University of Medicine and Pharmacy, Timisoara, Romania

²"Louis Turcanu" Clinical Emergency Hospital for Children, Timisoara, Romania

E-mail: dr.fiucupaun@yahoo.com, alinababeu@yahoo.com, damen.n.sebastian@gmail.com, boiaeugen@yahoo.com,

The abdominal ultrasound confirms, thickening of the gallbladder walls of 0.5 cm, aspect of biliary lithiasis with acute cholecystitis. Retro-urinary bladder transsonic-oval image of 7 / 3.5 cm raises the suspicion of collection. The gynecological consultation identifies an infantile uterus, with normal ovaries. The chest radiography was normal.

The abdominal-pelvic CT, native and with contrast, shows in the second segment from the left hepatic lobe a tumor with multiple-lobes outline of 4/3.8 cm, with content

of dense fluid type. Gall bladder with thickened wall, with limestone debris sediment.

At abdominal-pelvic level, collection with para-fluid densities of about 22 HU, dense proteinaceous increased content, bordered by a peripheral wall with capture of contrast substance, with sinuous distribution through the intestine loops, going from the bottom of the Douglas pouch up under to the lower pole of the right hepatic lobe (Figure 1).

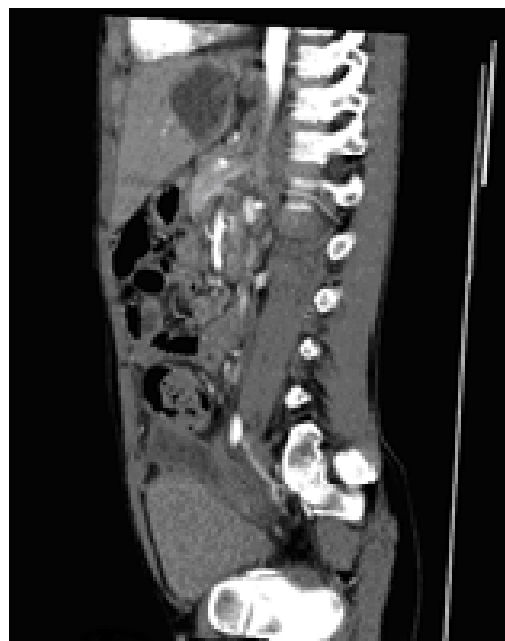
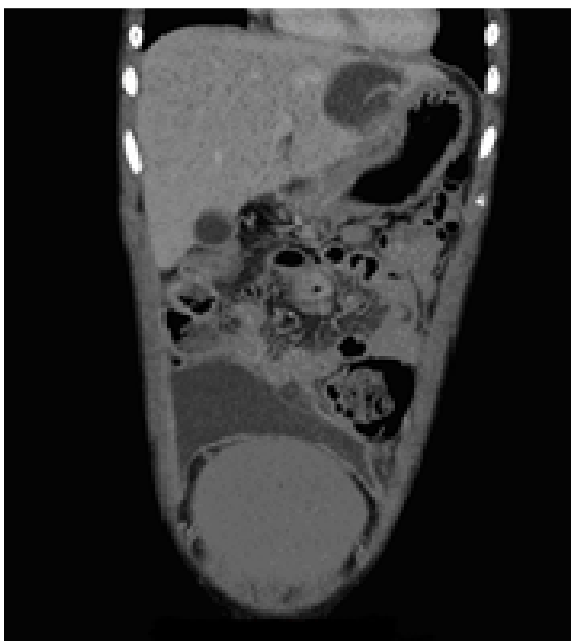


Fig 1. LHS ruptured hydatid cyst with intra-peritoneal adhesions between organs.

Corroborating the clinical data with laboratory, it is suspected the existence of a positive diagnosis of hepatic HC ruptured and discharged into the peritoneal cavity, biliary lithiasis with acute cholecystitis. The biological confirmation of the parasitosis is brought during hospitalization by ELISA test. Surgical exploration showed a large ruptured HC of the left lobe of the liver, with multiple daughter cysts in the peritoneal cavity.

Pre-operative therapy consisted of antibiotics Tazocin 2.25g IV 4x1 bottle / day, 200mg Albendazole 2x1 tablet / day, Paracetamol 3x35 ml bottle 100 ml IV/ day and hydro-electrolyte and acid-base rebalancing.

Surgical treatment consisted in median laparotomy with hepatic Lagrot procedure, cleaning the ruptured cystic and abdominal cavity with hypertonic saline 20% and its external drainage on silicone tube. A special attention was paid to the elimination of the vesiculae ficcae from the peritoneal cavity, and those fixed in the omentum removed en bloc by resection (Figure 2). Peritoneal fluid was collected for culture, the peritoneal cavity was washed with hypertonic saline 20% and the Douglas was externally drained with silicone tube.

The peritoneal cavity thorough inspection reveals a macroscopic phlegmonous appendix, reason why it was practiced the appendectomy.

The post-operative evolution of the case was without major complications. The patient was discharged 18 days after, in good condition. She received oral ALBENDAZOLE 15mg/kg body for three months. The post-operative follow-up period for the clinical and ultrasound re-evaluation took place at intervals of 3-6 months, not recording peritoneal recurrences. The hepatic cavity disappearance was found in ultrasound examination in 3 months after the hospital discharge and of the biliary lithiasis after 6 months.

Discussions

The complications of the hepatic HC in children are rare, represented by its rupture or fissure in the ducts or peritoneal cavity, over-infection or compression on the adjacent organs [10]. Its incidence varies in the literature from 1.75 to 8.6% [11] [12] [13].

The rupture and seeding of the peritoneal cavity occurs most frequently in the large liver cysts, after a trauma, or when conducting a corticalisation of the cyst on the underside of the liver. In contrast with Grozavu opinions, we recommend medical treatment as first therapeutic step because it doesn't lead to serious complications [14].

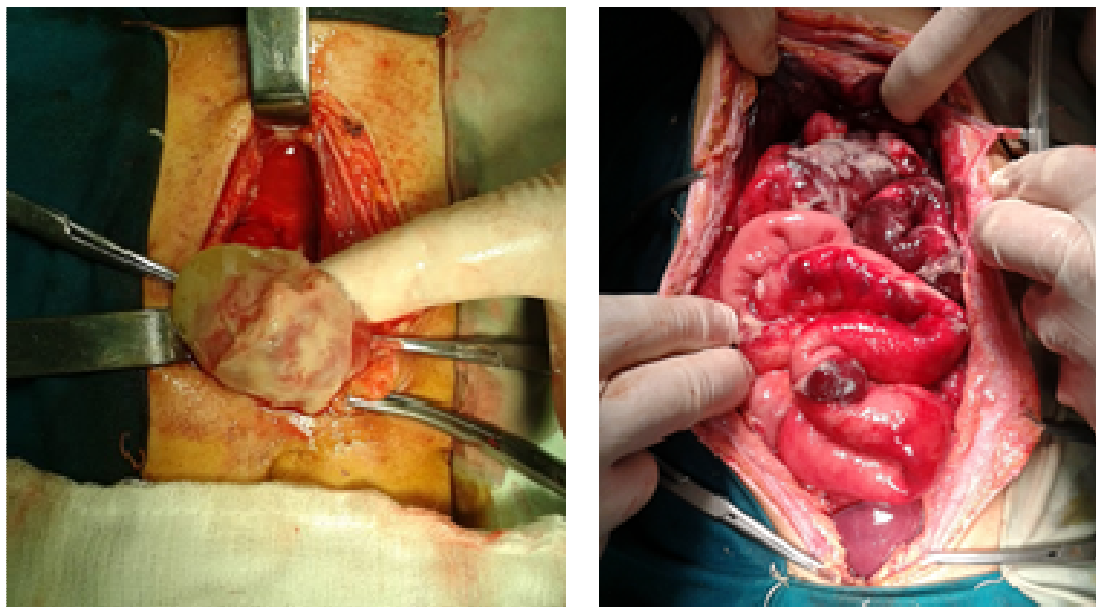


Fig 2. Hydatid cyst erupted in the peritoneal cavity vesicula ficae.

The hepatic HC, ruptured and emptied of its contents into the peritoneal cavity represents an immediate medical-surgical emergency, with the patient possible onset of anaphylactic shock or a moderate peritoneal irritation phenomena with secondary hydatidosis, without identifying the exact moment of rupture [15].

The clinical picture of this complication is influenced very much by the location and the size of the cyst, by the compression of the abdominal cavity organs. This picture is suitable to a differential diagnosis with acute appendicitis, peritonitis, or tumoral formation [16].

Most frequently, the rupture in the peritoneum is symptomatic, with constant abdominal symptoms [17] (abdominal pain, vomiting, abdominal tenderness and/or rebound), as well as marked allergic symptoms in up to 25% of the cases [13] (cutaneous rash, urticaria, anaphylactic shock).

We don't encountered anaphylactic shock as other authors reported (18).

Our intraperitoneal rupture was missed, due to absence of abdominal or anaphylactic complications, and due to the presence of diarrhea, which explains the late diagnosis, in contrast to other author's opinions [19] but not so late-until few weeks after the rupture. We discourage the wait and see tactics, in order to avoid any other possible complications, and we recommend immediate surgery. Peritoneal hydatidosis comprises 10–16% of intra-abdominal hydatid disease [20]. It mainly occur secondary to rupture of a hepatic or splenic cyst either spontaneously or accidentally during surgery [21].

Primary peritoneal hydatidosis accounts for less than 2% of intra-abdominal hydatidosis [22]. In our experience we do not encountered a primary peritoneal hydatidosis as Hegde N, et al. did [23].

Long-term monitoring is preferred but is difficult due to the nomadic lifestyle of the family and sometimes population migration

Conclusions

Patients with this complication are often operated for other conditions, such as acute appendicitis, peritonitis, acute cholecystitis or abdominal tumours.

The diagnosis and treatment of this complication is difficult and requires the existence of a medical team (radiologist, surgeon, anesthesiologist) working together in order to put the correct diagnosis and to succeed the surgery.

The surgical treatment targets both the host organ of the erupted cyst, as well as to improve the secondary peritoneal outbreaks and its lavage with hypertonic saline of 20%.

The post-operative follow-up interval in our case is quite low, but still, given that the rate of increase in size of the parasite depends also on the resistance of the surrounding tissues (minimum resistance in free peritoneum), a possible secondary hydatidosis could be evidenced by ultrasound or CT within 6 months after the onset.

Also, based on the presented experience, we consider that the peritoneal cleaning with hypertonic saline and the early and prolonged prophylaxis with anthelmintics may be salutary in order to avoid secondary echinococosis.

The diagnosis of hydatid cyst must be considered especially in endemic regions with a history of rearing livestock or owning pets, whenever a cystic mass is felt in the abdominal cavity [23].

References

1. Muller R, Wakelin D, Worms and Human Disease, 2nd ed. Wallingford, Oxon, UK: CABI Publishing, 2002
2. C. Smarandache, D. Căndeă, E. Kulcsar, R. Elefterescu, A. Șanta, C. Marosin: Hydatid cyst in children- Journal of Romanian Parasitology, vol.VI, nr.1-2; 1996
3. Cummings H, Rodriguez-Sosa M, Satoskar AR. Hydatid Disease. In : Satoskar AR, Simon GL, Hotez PJ, Tsuji M (eds) Medical Parasitology Austin, TX: Landes Bioscience, 2009, pages 146-152
4. Moldovan R., Neghina A.M., Calma C.L., Marincu I., Negina R. Human Cystic echinococcosis in two south-western and central western Roumanian counties: A 7-year epidemiological and clinical overview. Acta Tropica vol 121, Issue 1, January 2012; pages 26-29
5. Daliborca Cristina Vlad, Adriana Maria Neghina, Victor Dumitrascu, Iosif Marincu, Raul Neghina and Crenguta Livia Calma Cystic echinococcosis in Children and Adults: A 7-year Comparative Study in Western Romania
6. Brunetti E, Gulizia R, Garlaschelli AL: Cystic echinococcosis of the liver associated with repeated international travels to endemic areas. J Travel Med 2005 Jul-Aug; 12(4): 225-8[Medline
7. Ronald Barbosa, Ahmed Mahmoud, Nathaniel Matolo, Sheela Kapre, Gigant Hydatid Cystic Liver Disease: A Challenging Problem for Western Surgeons-Surgical Rounds, Issue: May 2006.
8. Schantz PM, Kern P, Brunetti E, Echinococcosis In: Tropical Infectious Diseases: Principles, Pathogens & Practice, 3 ed. Guerrant RL, Walker DH, Weller PF (eds.). Philadelphia: Saunders Elsevier, 2011, pages 824-838
9. Tantawy IM, Hydatid Cystic in Children. Ann Pediatr Surg 2010; 6 (2); 98-104
10. Irinel Popescu, C-tin Ciuce, Coordonator C. Sabetay Surgery disertation, volume III, ed. A II-a, Chirurgie Pediatrica/Pediatric Surgery. Editura Academia Romane Bucuresti 2014 ISBN 978-973-27-2185-8 pag. 253-362
11. Sözüer EM, Engin OK, Arslan M: The perforation problem in hydatid disease. Am J Trop Med Hyg 2002, 66(5):575–577.
12. Djuricic SM, Grebeldinger S, Kafka DI, Djan I, Vukadin M, Vasilievic ZV, Cystic echinococcosis in children – The seventeen year experience of of two large medical centers in Serbia. Parasitol Int. 2010; 59:257-261
13. Beyrouti MI, Beyrouti R, Abbes I, Kharrat M, Ben Amar M, Frikha F, Slim E, Walid G, Mohamed C, Ali G: Rupture aiguë du kyste hydatique dans le péritoine: À propos de 17 observations. Presse Med 2004, 33(6):378–84.
14. Gunay K, Taviloglu K, Berber E, Ertekin C: Traumatic rupture of hydatid cysts: a 12-year experience from an endemic region. J Trauma 1999,46(1):164–167
15. Grozavu C, Iliac M, Pantile D. Multivisceral Echinococcosis: Concept, Diagnosis, Management Chirurgia (2014) 109: 758-768 No. 6, November – December]
16. Dziri C, Haonet K, Fingerhut A, Treatment of hydatid cyst of the liver: where is the evidence? World J Surg 2004; 28 (8):731
17. Ozturk G, Aydinli B, Yildirgan MI, Basoglu M, Atamanalp SS, Polat KY, Alper F, Guvendi B, Akcay MN, Oren D: Posttraumatic free intra-peritoneal rupture of liver cystic echinococcosis: a case series and review of literature. Am J Surg 2007, 194(3):313–6
18. Derici H, Tansug T, Reyhan E, Bozdog AD, Nazli O: Acute intra-peritoneal rupture of hydatid cysts. World J Surg 2006, 30(10):1879–1883.discussion 84–85
19. Majbar AM, Mehdi Aalala, Mouna Elalaoui, Farid Sabbah, Mohamed Raiss, Abdelmalek Hrra and Mohamed Ahallat Asymptomatic intra-peritoneal rupture of hydatid cyst of the liver: case report BMC Research Notes 20147:114 DOI: 10.1186/1756-0500-7-114
20. Iuliano L, Gurgo A, Poletini E, et al. Musculoskeletal and adipose tissue hydatidosis based on the iatrogenic spreading of cystic fluid during surgery: report of a case. Surg Today 2000;30:947–9.
21. Yuksel M, Demirpolat G, Sever A, et al. Hydatid disease involving some rare locations in the body: a pictorial essay. Korean J Radiol 2007;8:531–40
22. Khuroo MS. Hydatid disease: current status and recent advances. Ann Saudi Med 2002;22:56–64
23. Nishchit Hegde and Bharati Hiremath. Case Report, Primary peritoneal hydatidosis. BMJ Case Rep. 2013; 2013: bcr2013200435. Published online 2013 Aug 2. doi: 10.1136/bcr-2013-200435PMCID: PMC3762414.

Correspondence to:

Fuicu Paun
 Strada Iosif Nemoianu 2,
 Timisoara,
 Romania,
 E-mail: dr.fuicupaun@yahoo.com