HEPATIC CYSTIC ECHINOCOCCOsis STUDIED IN A FAMILY GROUP

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Abstract

Introduction. Infection with Echinococcus granulosus is a disease known since around 1550 BC. In 1786, Batsch was the first to discover the etiological agent of the disease. The main intra-abdominal location of the hydatid cyst is the liver. The way of transmission of the disease leads to a possible infection of several members of a family. Materials and methods. In 2014, patient R.M. was admitted in the Surgery Clinic of Colentina Hospital, being diagnosed with liver hydatid cyst. Lagrot operation was performed, followed, postoperatively, by ERCP with endoscopic sphincterotomy. Investigating patient’s heredo-collateral antecedents, we discovered that 3 of her 4 children had been diagnosed and treated for liver hydatid cyst. In the fourth child and in patient’s husband, the absence of the disease was documented. Results and discussion. Both the patient treated in Colentina Hospital and her children had a favorable post-treatment evolution, as confirmed by the latest evaluation performed in 2015. Conclusions. Infection with Echinococcus granulosus in one of the family members raises suspicion of affection in the other members too, in order to identify all possible cases.

Keywords: hepatic cystic echinococcosis, familial disease, children, infections.

1. INTRODUCTION

Parasitic infection with Echinococcus granulosus is a disease known since antiquity, being mentioned by Egyptians in Eber’s papyrus, around 1550 BC. In 1786, Batsch discovered the etiological agent of the disease [1]. The endemic areas for hydatid disease are the Mediterranean Basin, North and East Africa, South America, Australia, New Zealand, Middle East, China, India [2-4]. Although Romania is not among these areas, studies have identified both an increase in the incidence of hydatid disease, reaching 5-6 cases per 100,000 inhabitants / year, as well as a high rate of surgical interventions used to treat this pathology [5-7]. Hydatic disease occurs in humans as a result of either direct contact with an infected dog (definitive host), or by ingestion of food contaminated with the feces of such a dog [8]. The liver is the main localization of the hydatid cyst, followed, as to frequency, by lungs, spleen, kidneys and brain [9-11]. Unless the hydatid cyst is large or complicated, the diagnosis is accidental, due to a routine abdominal ultrasound, or established for other pathology [12]. In uncomplicated forms of the disease, patients can accuse discomfort or pain in the right hypochondrium, abdominal meteorism, fatigue, nausea, the symptoms depending on the location, number and size of cysts. Diagnosis is determined by imaging and laboratory investigations, the treatment involving either a drug treatment, its association with surgical treatment or, more recently, minimally invasive techniques [13]. The WHO-IWGE ultrasound classification includes the hepatic hydatid cysts into one of the six CL-CE5 categories and, on the other hand, it allows the choice of the optimal therapeutic tactics. The mode of transmission of the disease favors the possibility that several members of a family who come into contact with infected animals are affected, patients being unaware of
the presence of the disease, due to reduced and unspecified symptomatology.

2. MATERIALS AND METHODS

We discuss the case of the R. family, from Buzau County, consisting of 6 members: mother, R.M. aged 38, father, R.C., aged 40, child I, RA, female, 17 year-old, child II, RD, female, 16 year-old, third child, male, RA, aged 11 years and child IV, RA, female, aged 9. The mother was admitted in the Colentina Hospital Surgery Clinic in 2014, after an initial evaluation in the Clinic of Parasitology, where she was diagnosed with giant hydatid cyst located in the right liver lobe, after abdominal ultrasound was performed and ELISA test was positive for *Echinococcus granulosus* antibodies. Reasons for admission were pain in the right hypochondrium and nausea, the symptoms starting a month before. From the heredo-collateral history we identified the presence of *Echinococcus granulosus* liver disease, documented imagistically and serologically, in 3 of the 4 children of the patient: child I and II, who received surgical and pre- and post-operative medical treatment with Albendazole, and child III, who received only Albendazole. All children had been diagnosed with hepatic hydatid cyst in 2010, child I presenting a hydatid cyst of stage C3a with a size of 5.5/3.4 cm located in the right liver lobe (Fig. 1), the second child - 2 CE1 cysts with a size of 4.7/4.9 cm and 3.3/3.1 cm, respectively, localized in the right hepatic lobe (Fig. 2), and child III presenting a CE1 cyst of approximately 2.8/1.5 cm, localized in the left liver lobe (Fig. 3). Child IV and patient’s husband, examined by ultrasound, pulmonary radiography and ELISA for Ac anti-*Echinococcus granulosus*, were declared non-carriers of the hydatid disease. Mother’s blood samples at her admission showed normal values, except: TGP = 57 IU/L, GGT = 123 IU/L, alkaline phosphatase = 148 IU/L, fibrinogen = 865 mg/dl. The patient was re-evaluated through abdominal ultrasound at the time of admission, the result showing a transsonic image in the hepatic segment VII, approximately 13 cm in diameter, with another image within it, about 3 cm in diameter, biliary sludge in the cholecyst, without further intraabdominal echographic changes (Figs. 4 and 5).
Pulmonary radiography did not detect pathological changes. Spirometry has identified normal values of respiratory parameters. Superior digestive endoscopy performed preoperatively revealed hyperremic gastritis, more pronounced in the anthral region, and large duodenal papilla protruding into the lumen. Abdominal CT examination identified hepatomegaly with cranio-caudal diameter of the right hepatic lobe of 20 cm, cystic wall-shaped cavity which occupies almost completely segments VIII, VII and IV A with axial diameters of 15/11 cm and a cranio-caudal diameter of 11
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cm, with the lower pole in segment V, with
decolated membrane, daughter cells and areas of
bleeding in the upper parietal pole, containing
non-iodophilic clear liquid, the mass affecting
the vascular structures, especially the
suprahepatic veins and portal veins, as well as
the left hepatic biliary duct, causing IHBD
dilatation without PBD dilatation, cholecyst
containing a 0.5 cm diameter stone and non-
obstructive left kidney stone. Note that, at the
time of admission to the Surgery Clinic, the
patient was under medical treatment with
Albendazole, initiated 2 weeks ago. Surgical
intervention under general anesthesia with OTI
was performed; intraoperatively a voluminous
cyst (approximately 15 cm in diameter), without
expression on the visceral face of the liver,
occupying IV, V, VII, VIII segments with multiple
adhesions to the diaphragm, was discovered.
Adhesiolysis, cholecystectomy, cystectomy with
partial peri-cystectomy and double drainage of
the remaining cavity, after cyst inactivation with
hypertonic serum, subfrenal right and subhepatic
drainage were performed. Postoperative
evolution was slowly favorable, except for a bile
stream of approximately 400-500 ml / 24 hours
through the drainage tubes from the remaining
cystic cavity, and an infection with Enterobacter
in this drainage. In order to reduce the external
biliary drainage, ERCP with endoscopic
sphincterotomy was performed, and antibiotic
treatment with Amikacin and Colistin was
administered against the Enterobacter infection,
according to the antibiogram.

3. RESULTS

The first child, in whom the postoperative
ultrasound detected residual cavity and residual
hepatic hydatid cyst, received continuous
treatment with Albendazole for 9 months,
followed by discontinuous Albendazole 90-day
cures with 2 week pauses between them. The last
evaluation, in September 2015, showed the
absence of relapse (Fig. 6). The second child
received drug treatment for 5 months
postoperatively, the last evaluation in September
2015 demonstrating the absence of relapse (Fig.
7). Child III received discontinuous drug
treatment for 8 months with 3 month breaks. The
last evaluation, performed in September 2015,
showed the absence of the disease (Fig. 8). The
mother, patient R.M., discharged from the
Surgery Clinic 42 days after surgery, received 6
months of treatment with Albendazole
postoperatively and was evaluated in September
2015, when no recurrence was found.

4. DISCUSSION

The presence of several cases of hepatic
echinococcosis within the same family is
explained by the fact that all patients were
exposed to the same risk factors: contact with
possibly infected dogs, water consumption and
potentially contaminated food with Echinococcus
granulosus eggs, and lack of a proper hygiene. At
the same time, patients come from an endemic
area known for sheep breeding. The increasing
incidence of the hydatid disease could be an
argument in favor of extending investigations to
the members of a family in which a disease case
occurred, even if we are not talking about an
endemic area, as also suggested by Eda Karadagli
et al., which supports the idea of family screening,
especially in risk areas, but not only (14). An
efficient evaluation of all family members at the
time of the first case of hepatic hydatid cyst
would have provided the possibility of a rapid
diagnosis of the disease in R.M. (mother). Except
one of the cases (child III), in which healing was
achieved only by administering the specific
medication, all the other family members
received surgical treatment, associated with
medication. The surgical technique chosen in the
case of the mother was dictated by the fact that
the cystic formation had no expression on the
visceral hepatic face, but on the diaphragmatic
one, a level at which it could be approached
without major risks. Alongwith the imaging and
serological methods used in hydatidosis
screening, good information on the risk of
developing disease in endemic areas and on
prevention methods of the disease provides the
premises for better control of disease spreading
within a community.
5. CONCLUSIONS

Once a case of hydatidosis occurs within a family, it is justified to carefully evaluate the other members of the family, both through a detailed anamnesis and through imaging and serological methods, thus offering the possibility of identifying all possible cases. Early diagnosis of echinococcus infection allows the establishment of drug therapy that offers the chance of complete healing and reduces the number of cases requiring surgical treatment to achieve this goal.

References