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Case Report

# Pulmonary, Splenic and Hepatic Hydatidosis : An Unusual Multivisceral Location in Children : A Case Report

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## Abstract

Hydatid cyst is a parasitosis due to the development of the larval form of Echinococcus granulosus which can infest all organs. The pulmonary location comes in 1st position followed by the liver and the spleen. The discovery is generally fortuitous on ultrasound. Hydatid serology guides the diagnosis. Surgical treatment, whether conservative or radical, gives good results. We report the case of a 10-year-old child from Morocco, without any history, who consulted us for a dry cough associated with a medium-sized hemoptysis, a thoracic pain of the right hemithorax and a heaviness of the right hypochondrium revealing a hepatic and splenic hydatid cyst. Although splenic and hepatic hydatidosis remains rare in the pediatric population, this observation serves to emphasize this pathology as well as other differential diagnoses.

## Introduction

Human echinococcosis is a zoonosis, caused by parasites, the tapeworms of the genus Echinococcus. There are essentially two forms of echinococcosis, cystic echinococcosis or hydatidosis, due to Echinicoccus granulosus, and alveolar echinococcosis, due to E. multilocularis. Hydatidosis or cystic echinococcosis is a public health problem in livestock areas of developing countries, particularly in the Mediterranean basin, North Africa, Latin America, Australia, New Zealand, China and Central Europe. Overall, exposure to food and water contaminated with feces from an infected host or poor hygiene in areas of infestation can cause this disease. The splenic localization comes in 3rd position after the liver and the lungs, but any organ can be affected, with simultaneous localization to one or more viscera. The presentation of primary dissemination through multi-organ involvement is very rare, especially in the pediatric population. In the absence of truly effective medical treatment, splenic hydatidosis often leads to surgery. We report the case of a 10 year old child with splenic hydatidosis revealed by a medium-sized hemoptysis associated with a dry cough admitted to the pediatric ward of the University Hospital of Marrakech, after free and informed consent by the parents.

**Keywords:** Child; Hydatid cyst; Hydatidosis; Liver; Spleen; Splenectomy

### Observation

The child was El.A., 10 years old, with no particular pathological history, vaccinated up to date according to the national immunization program. He presented for four days with a dry cough associated with four episodes of moderate hemoptysis, chest pain in the right hemithorax and heaviness in the right hypochondrium, all evolving in a context of uncalculated fever and conservation of the general condition. On clinical examination, the patient was conscious, polypneic at 25cpm, normocardial,

apyretic at 36.6°C, SpO2 at 98% on room air, with normal staturo-ponderal development. The pleuropulmonary examination showed right basithoracic crackling rales without signs of respiratory struggle. Abdominal palpation revealed tenderness in the left hypochondrium. In front of this presentation, three diagnoses were evoked: a hydatid cyst, pulmonary tuberculosis and pneumonia. A chest radiograph showed an alveolar opacity of the middle lobe (Figure 1).

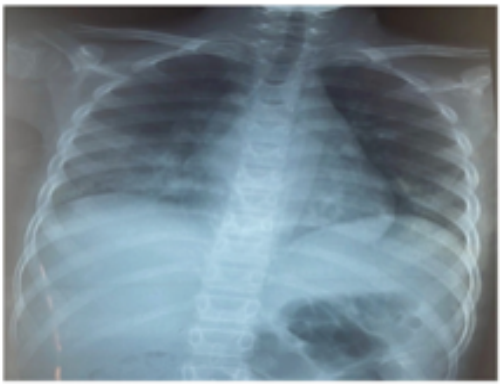

**Figure 1:** Frontal chest radiograph showing an alveolar-type opacity occupying the middle lobe in a child El.A.

An abdominal ultrasound was performed, the aspect of which was in favor of hepatic hydatid cysts in segments VII (13.5*3.1mm) (Figure 3), II (18*15.1mm) (Figure 4) and I (16.1*4. 7mm) (Figure 5) and splenic type I of the GHARBI classification (Figure 2), oval shape, well limited, anechogenic content, generating a posterior enhancement, non vascularized on color Doppler measuring 53*52mm.

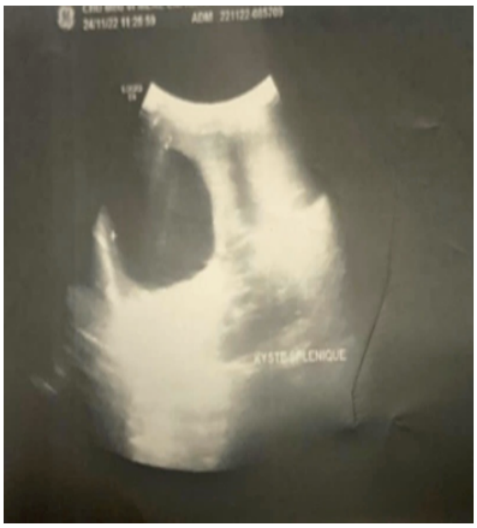

**Figure 2:** Anechoic splenic cystic formation, roughly rounded in shape with a thin wall and no partitions or endocystic vegetations generating a posterior reinforcement.

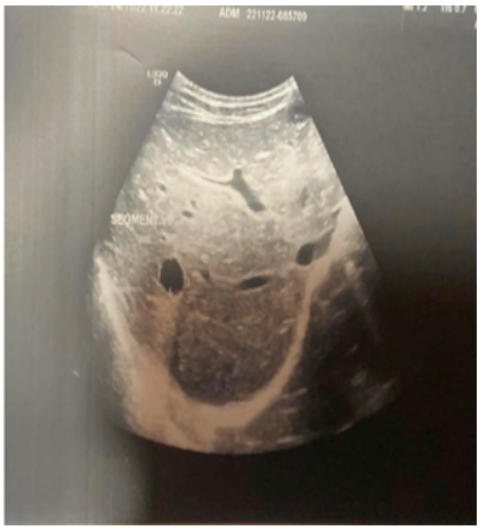

**Figure 3:** Cystic formation of segment VII measuring 13.5*13.1mm.

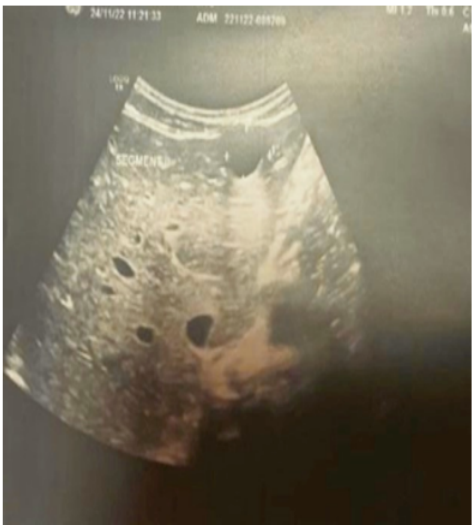

**Figure 4:** Cystic formation of segment II measuring18*15.1mm.

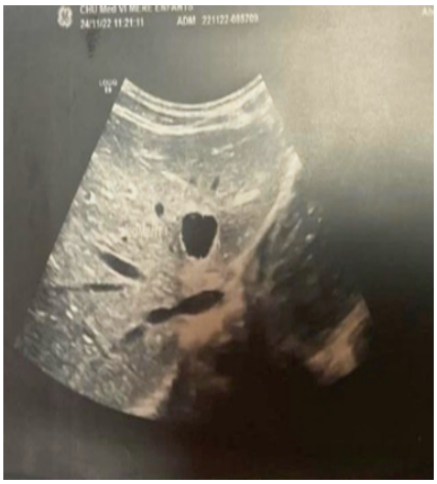

**Figure 5:** Cystic formation of segment I measuring 16.1*4.7mm.

A thoracoabdominal CT scan performed showed hepatic and splenic hydatid cysts (Figure 6).

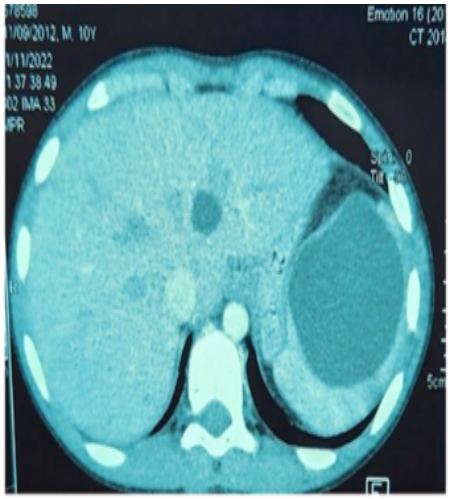

**Figure 6** : Splenic hydatid cysts measuring 5cm and hepatic segment II measuring 18*15.1mm.

As part of the etiological workup, a tuberculosis workup including three sputum BKs, a TST and an Xpert test was negative. An infectious workup was performed: CBC: WBC= 13400 PNN=9220 Lymphocytes=2730 Eosinophils=700 Hb=10.4g/dl Platelets= 350,000. A hydatid serology was performed and came back positive at titre 1/2560. In view of this unusual and multivisceral presentation, an antiparasitic treatment based on imidazole Albendazole 15mg/kg/d for 03 months was instituted and then he was referred to the pediatric surgery department for surgical management which consisted of a puncture of the splenic cyst with aspiration of the contents followed by the exteriorization of the proligeral membrane with a puncture, aspiration and extraction of the hepatic cysts of the left lobe The postoperative course was simple (Figures 7a,7b).

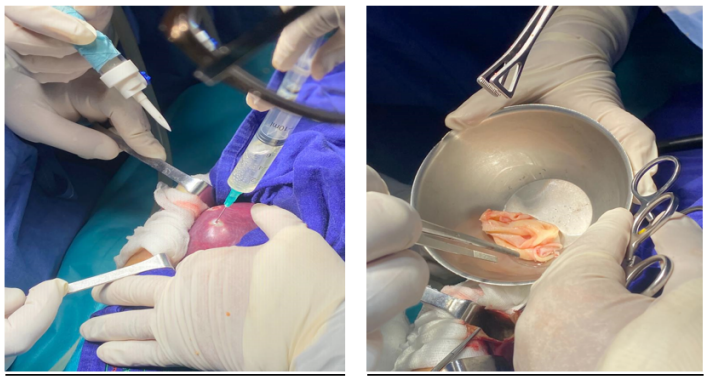

**Figure 7:** A: intraoperative view of splenic hydatid cyst puncture-aspiration. B: postoperative view of splenic hydatid cyst.

## Discussion

Hydatidosis is a helminthiasis caused by the development in humans of the larval form of Echinococcus granulosus. The classical cycle is the domestic cycle: dog (HD) - sheep (HI). Man is an accidental host: it is a parasite dead end. The embryonated eggs, eliminated in the external environment with the dog's faeces, are ingested by man, penetrate the digestive wall, reach the liver via the portal system, sometimes pass through the supra-hepatic veins and reach the lungs. More rarely, the localization can be done in any point of the organism by the general circulation, the splenic localization thus comes in 3rd position. Once in the viscera, the embryo turns into a hydatid larva. The cycle is closed when the dog devours the viscera of a parasitized herbivore such as sheep [1,2].

The case we report is exceptional because of the multiplicity of organs affected and the very young age of the patient. In children, male involvement is greater than female involvement. This can be explained by the fact that boys are more promiscuous with the dog, as they spend more time playing outside, and are therefore more exposed to embryophores [3]. This male predominance is reported by many authors, with sex ratios of 1.12 in Sayir et al [4], 1.27 in Hafsa et al [5]. The sex of our patient is part of this generalization of male predominance. The age of our patient is 10 years, which is in line with the results of other series where the majority of children are older than 5 years [6]. For some authors, the age of discovery is earlier: Lagardère et al [6] described a case at 9 months, C. Hafsa [5] described a case at 18 months, and M. Bouskraoui et al [7] reported a case of pulmonary, splenic and hepatic hydatidosis in a 23 month old infant. In the pediatric population, cysts develop more rapidly in compressible organs such as the lungs and brain,

which may explain the relatively high incidence of the disease in these organs in children [8]. While in adults, liver involvement is more common than lung involvement, in children the reverse is true. In fact, our patient has liver and spleen cysts but also a cystic cavity in the lung, indicating a rupture of a pulmonary cyst, which explains the episodes of hemoptysis but also the abdominal pain.

A study of 10 children aged 2-12 years concluded that children are more likely to have an atypical presentation of hydatid cyst disease than adults [9]. Sometimes it is not possible to explain the pathogenesis when there is an atypical location of HD, but when cysts occur in the peritoneal cavity, as in our patient's case, the cause may be related to the unsuspected presence of a very small liver cyst that ruptured spontaneously and infected the spleen [10,11].

We find that children present with vague symptoms including mild fever, non-specific pain, cough and abdominal heaviness. In its splenic localization, hydatid cyst is characterized by a late evolution; on average 3 to 4 years [12]. It therefore remains asymptomatic for a long time, revealing itself by heaviness, pain and a sensation of a mass in the left hypochondrium. All these symptoms were noted in our patient, so the finding in the patient of a dry cough, hemoptysis but especially chest pain in the left hypochondrium made the lead to cystic echinococcosis prevail. Like the case presented, the circumstances of discovery of hydatid cysts are often fortuitous on the occasion of abdominal ultrasound. As previously mentioned, the diagnosis is essentially based on imaging, the key examination being ultrasound, which in our case revealed hepatic and splenic hydatid cysts TYPE I of the GHARBI classification. For a long time this classification was used, having the merit of being the oldest and most widely used, but it has now been replaced by the WHO classification [13], which has the advantage of correlating the ultrasonographic aspect with the evolution of the parasite, which can therefore allow us to start considering therapeutic management as soon as the ultrasound is done. The GHARBI 1 or CE1 stage (New WHO classification) corresponds to an Active EK (cystic echinococcosis) due to a proligeral membrane that is perfectly attached to the wall. The cyst presents as a simple cyst with an anechogenic content or we can sometimes detect fine echoes that correspond to hydatid sands. What is very important is the demonstration of a double contour aspect in the wall which is pathognomonic for the diagnosis of the case. Our patient's CT scan showed cysts with intracystic calcifications of the spleen; (Figure 2).

Hydatid serology plays a secondary role in the diagnosis but is an essential step in the orientation of the diagnosis in 80 to 95% of hepatic localizations and in 40 to 65% of pulmonary localizations [14]. Serology can confirm the suspected hydatid etiology in the light of the results of radio-clinical investigations and can also be used to monitor the efficacy of the treatment via the serum antibody level. Indeed, the The persistence of a high level of antibodies beyond a period of 12 to 24 months suggests a failure of the treatment. Our patient's hydatid serology was also positive at titer 1/2560 (threshold of positivity 1/320) [15] confirming the diagnosis of splenic and hepatic KH. However, in some cases, cysts may be serologically negative but this does not necessarily exclude the diagnosis of hydatidosis. Indeed, in a retrospective study of 114 cases of pulmonary hydatid cysts, hydatid serology was performed in 73% of cases and only 54% of the tests were positive [16]. It is important to note that the value of direct specific diagnosis remains very limited, as it is only possible to search for the pathogen in intraoperative cyst samples. Indeed, it is formally contraindicated to perform a cyst puncture, as there would be a risk of dissemination with secondary echinococcosis, and of anaphylactic shock [17].

Medical treatment with imidazoles can be prescribed for multi-visceral forms even if the results are insufficient [18]. Benzimidazole derivatives are relatively effective against hydatidosis. Mebendazole (Vermoxt) was tested in the 1970s. In the early 1980s, albendazole (Zentelt) proved to be far superior [19,20]. We assume that medical treatment results in a cure in 30% of cases, improvement in 40-50% and no response in 20-30% [20].

The response rate with albendazole is 75%; it is less than 50% with mebendazole. The optimal duration should be three or four 28-day courses, separated by 14-day intervals. Some authors recommend continuous treatments of 3 to 6 months [21].

Thus, we prescribed to our patient a dose of 15mg/kg/d for 3 months of Albendazole before surgery. Surgery remains the solution of choice, especially in complicated forms. Indeed, the treatment of hepatic and splenic KH is essentially surgical. In our case, the treatment of the liver (cysts of segments 1, 2 and 7) and of the spleen consisted of removal of the cysts by median supra-umbilical laparotomy, which was rapid and allowed a good exploration of the abdominal cavity. It perfectly exposed the cysts of the left liver but was somewhat limited on the right locations, in particular those of the posterior sector. During the intraoperative exploration we found a cyst in the splenic area and two cysts visible in the left lobe of the liver and a palpable cyst in the intra parenchymal area of the right lobe which was not accessible. The liver cysts in the left lobe were punctured, aspirated and then removed. The cyst of the right lobe was inaccessible because of the nature of the approach chosen, which is limited to the right locations. We therefore decided to leave this last cyst intact and to count on a favourable evolution thanks to the medical treatment. The splenic cyst was punctured on its dome through a large trocar and emptied by strong suction. The puncture brought back a clear fluid. The opening of the peri-cyst then allowed evacuation of the

hydatid debris and of the proligeral membrane.

Total splenectomy has the advantage of removing the parasitized organ and avoiding recurrence. But it is difficult to perform in case of cysto-visceral adhesions. The mortality rate reported in the literature ranges from 3.7 to 22.5%, while the morbidity is 21 to 25% [22]. Serious complications may be encountered, especially in children [23]. Therefore, total splenectomy seems less legitimate, especially in the presence of benign pathology. Conservative surgical techniques are therefore to be encouraged, although they have their own complications. RDS has the advantage of being a benign procedure, with little bleeding, and is almost always feasible, as long as the KH is accessible on the surface of the organ treated [24,25] Its disadvantage is that it leaves the pericyst in place, which may be the site of a residual cavity and postoperative infection. With regard to lung involvement and possible ruptured cysts at this level, we wondered whether an exploration of the residual cavity might be necessary to look for possible cysts at this level.

## Conclusion

Hydatidosis is a public health problem in developing livestock areas. Its prognosis has been modified by new therapeutic possibilities, but remains insufficient as long as prophylactic measures do not accompany the progression of medical and surgical solutions. The hydatid disease could only disappear thanks to strict preventive measures which cannot be implemented without the improvement of the standard of living of the populations. These measures start with health education of the population in endemic areas, veterinary control of livestock slaughter, slaughter of stray dogs with census and deworming of domestic dogs.

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