



A hydatid cyst in mesentery complicated with appendicitis in nine-year-old child

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ABSTRACT

Hydatid disease is a parasite disease caused by *Echinococcus granulosus*. It often forms in the liver and lungs and rarely forms in other parts like the mesentery as in our case. This rare location of the cyst creates a unique and challenging condition. The patient in our case complained of abdominal pain, diarrhea, nausea, and vomiting. The radiographic and laboratory investigations support the diagnosis of appendicitis, so the appendix was excised and the patient was discharged after he got better. But he returned to the hospital complaining of abdominal pain and vomiting the next day. The X-Ray and laboratory values did not help us in putting the diagnosis, so an investigative laparotomy was done. We found a hydatid cyst in the mesentery of the ileum. We excised it carefully and treated its cavity with a saline solution of 30%. The patient was treated with albendazole for six months. The CT scan and MRI confirmed that it is a primary cyst. This case should clarify that hydatid cysts can form in unusual locations and lead to different symptoms and signs. And it can be complicated by other medical problems. So, the comprehensive examination is important to exclude such urgent and dangerous cases, particularly in endemic areas.

1. Introduction

Hydatid disease is a zoonosis disease caused by the cestode called *Echinococcus granulosus*. There are three other genus of *Echinococcus* that cause infection in human called: *Echinococcus multilocularis* (alveolar hydatid disease), *Echinococcus vogeli* and *Echinococcus oligarthus* (both causing polycystic hydatid disease) [1]. This disease occurs when humans eat the eggs of the Cestoda and digest the protective layer by enzymes in the gastrointestinal system, releasing the Oncospheres which transfer blood or lymph to different parts of the body. Even though the definitive host is dogs and cats, the larval stage infects the human as an intermediate host [2]. The prevalence of this infection is not known accurately in many regions, but in others, it is considered an endemic disease such as North Africa, the Mediterranean region, Eastern Europe, and the Middle and Far East [2,3]. The mortality rate for this disease in the medical literature is approx 4% [4]. The cysts can form in any part of the body. The most involved organ is the liver (59–75%), followed in frequency by the lungs (27%), bones, kidneys, and brain. It rarely consists in other locations such as the back, soft tissues, ovaries, heart, spleen, pancreas, peritoneal cavity, omentum, and mesentery [4]. The presence of cysts in these rare locations can create difficulties in diagnosis and treatment like in our case, which is complicated with appendicitis. We report this case to highlight one of the rarest causes of the acute abdomen which requires urgent treatment, especially in endemic regions.

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2. Case presentation

A nine-year-old boy came to the emergency department and complained of abdominal pain, diarrhea, nausea, and vomiting. The pain started in the morning in the umbilical region with severe mucoid diarrhea (about 10 times) and vomiting after any oral supply. The local doctor in the patient's area diagnosed the case as gastroenteritis and treated him with physiological serum, antiemetic, Metronidazole, and anti-inflammatory injections. The fever responded partially but the general situation did not get better. The physical examination in the hospital revealed generalized abdominal pain, increasing in the right iliac region with defense. The child was conscious and responsive, the pulse was palpable and the fever was slightly high. There was no pallor, bleeding, or bruising on the skin. Other systems were normal including the glandular, lymphatic, musculoskeletal, cardiovascular, and respiratory systems. The child was born by cesarean section without any need to stay in incubators, his nutrition in the first year depended on artificial feeding, and his vaccines were complete. Spiritual and psychomotor development was normal. There was no personal or familial illness. The ultrasound was done and it revealed that there was thickening in the walls of the appendix with a little amount of free liquid in the abdomen and pelvis, other findings were normal. The CBC was done and it revealed that there was an increase in the number of white blood cells (total number 10.6×10^3 UL, the neutrophils 9.2×10^3 UL (86.4%), the lymphocytes 1.1×10^3 UL (10.1%)). The red blood cells were 4.19×10^6 UL, the hemoglobin was 10.4 g/dl. HCT, MCV, MCH, and MCHC were all low. The electrolytes including Sodium, Potassium, Chlorine, and Calcium were normal. Urinalysis revealed the following values: Density 1.015, the color was yellow, pH 6.5, white blood cells 3–4, red blood cells 6–8, epithelial cells were positive, and oxalate crystals were positive. The case was diagnosed as acute appendicitis and the appropriate surgical approach was done. The excised appendix was inflamed and justified the symptoms, so we did not investigate the abdomen. The patient was discharged from the hospital the next day after checking that the patient's condition was good and stable. He developed severe vomiting and abdominal pain the next day and it was treated at the home without any improvement, so he came to the hospital. The ultrasound revealed dilatable pontoons and cystic formation with barriers in it behind the umbilical, measuring about 70 mm. The X-Ray in a standing position was done and showed gas-liquid levels (Figs. 1 and 2). The CBC was done and revealed elevated white blood cells, the other values were the same. The different findings were making the diagnosis very hard, so investigative laparotomy was done. We found a mass measuring about 60*100 mm at the expense of the mesentery of the ileum, away from the ileocecal valve about 600 mm. we treated it very carefully after surrounding it with sterile fabrics to turn out that it was a hydatid cyst. We excised the germinal wall and daughter cysts, treated its cavity with a saline solution of 30%, partially excised its wall, and closed it (Figs. 3–5). The patient was treated with albendazole for 6 months. The CT scan and MRI were done and showed that there was no any cyst in any part of the body.



Fig. 1. The X-Ray in a standing position shows gas-liquid levels.



Fig. 2. The X-Ray in a standing position shows dilatatable pontoons.

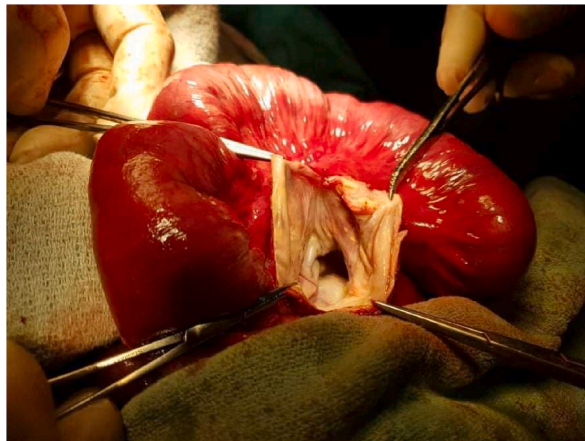


Fig. 3. Image during operation reveals the germinal membrane inside the mesentery.

3. Discussion

Hydatid disease is a parasite disease caused by *Echinococcus granulosus* [5]. It infects the humans as intermediate host and cats and dogs as the definitive host. It is an endemic disease in some areas of our country (Syria) while it is rare in others [2]. It can occur in any part of the body but the most influenced organs are the liver and lungs. The formation of hydatid cyst in other places like the mesentery is very rare and make the condition very hard to diagnose and treat [4]. The probability of a cyst appearing in the peri-



Fig. 4. Image during operation reveal the sutures after excising the daughter cysts.



Fig. 5. Image shows the daughter cysts.

toneal cavity including the omentum and mesentery is about 2% [6]. The cyst consists of many layers forming the wall. The outermost layer forms from the immune reaction of the host, the middle layer is the laminated membrane and the innermost layer is the germinal membrane which forms another cyst [4]. We can see these structures through histopathology which we do not need because of the descriptive appearance of the cyst in our patient (Figs. 3–5). We can recognize many types of cysts depending on the stage of development. Type I is a cystic mass composed of a regular round or oval wall and fluid inside it, with no other structures within the fluid. Type II is forming when matrix or daughter cysts appear inside the fluid like in our case. Type III forms when type II lesion consumes all the nourishing liquid and starts to starve and die, so we see calcifications in it, which is a distinctive component for this type [2,4]. We can differentiate these types from each other before excising through imaging including ultrasound imaging, CT scan, and MRI. After excising, we depend on histopathology or visual appearance. In our case, we have a cyst with daughter cysts inside it (Fig. 5) so it is type II. The distribution of the hydatid cysts can differentiate between adults and children. Some studies show that the most influenced organ in adults is the liver then the lungs, while in children the most influenced organ is the lungs then the liver. As well as the probability of cysts appearing in atypical locations in children is more frequent than in adults for unclear reasons [7]. There are two varieties of cysts that can be found in the mesentery: primary and secondary. The cysts are considered primary cysts when there is no other accompaniment cyst, which is a very rare case [1]. There are some hypotheses that explain the mechanisms of forming the cyst in our case. The first one supports the idea of hematogenous or lymphatic diffusion of the oncosphere through the intestinal wall to the mesentery either after passing the liver or without [1,8]. Another hypothesis is that a microscopic hydatid cyst wall may split, expelling the germinal membrane from its place without rupturing apart from the adventitia, forming a fibroid membrane around itself; the exposed cyst fixes to the serosa to keep its development. The new cyst is described as heterotypic primitive echinococcosis, and there is no evidence of rupture in the majority of cases [8]. The cysts are considered secondary cysts when they accompaniment with others in the liver. The liver can rupture spontaneously into the abdominal cavity and form a new cyst in it and its associated organs. In our case, the MRI and CT were done out of the hospital after the surgery and because of the hard situation we could not get them, but through social media, we contacted a doctor in the patient's area and he confirmed that there were not any findings inter the liver [6]. Based on the above the cyst in our patient either is a primary cyst which is a very scarce case or it is secondary to a very small or visceral hydatid cyst we cannot figure out through available imaging equipment. The symptoms in such cases are atypical and can vary broadly, but abdominal pain and distention are still the most common symptoms according to the study in the English literature which reported cases of the primary hydatid cysts in the peritoneal cavity, omental, and mesentery in the whole world during the last 20 years. Even though some patients are asymptomatic for a long time, others complain of large size and pressure effect [6]. In addition to the mentioned symptoms, the patient may suffer from one of the complications like rupture, cyst infection of the biliary tree, and anaphylactic shock [1]. The rupture of the cyst can occur in the biliary tree, peritoneal cavity, adjacent organ, or pleural cavity [5]. The first case that reported a traumatic rupture of a hydatid cyst in a mesenteric location was published in 2003 [9]. The rupture occurs in 50%–90% of cases. There are three types of rupture: contained, communicating, and direct. In contained rupture, the innermost layer separates from the outermost layer without any contact with the outer medium. In communicating rupture, the cysts rupture into nearby organs or structures such as a biliary tree, bronchial tree, or abdominal cavity. Intrabiliary rupture which is one of the most common and serious complications can lead to severe problems like biliary obstruction, Cholangitis, pancreatitis, cholecystitis, and septicemia. This condition often accompanies high morbidity and mortality. In direct rupture, the innermost and the outermost layers split and the cyst's contents are emptied into a closed cavity [4,10]. The causes of rupture include trauma, medical treatment, and endocyst degeneration and it can lead to acute abdomen [3]. The acute abdomen condition that the patient suffered from after excising the appendix may result from the surgical procedure which incited a rupture in the hydatid cyst and led to the mentioned symptoms. And in turn, small or even microscopic ruptures in the cyst and infiltration of the liquid to the peritoneal cavity may incite appendicitis. The infection of the hydatid cyst ranges from 11% to 27.1%. It results from communication between the cyst and the outer medium. The infection causes many symptoms and mortal conditions such as abdominal pain, fever, chills, jaundice, nausea, vomiting, and anemia. So any contamination during the appendectomy may lead to infection in the hydatid cyst [5]. The diagnosis of hydatid disease basically depends on imaging methods that differ according to the availability of equipment and the site of lesions. Ultrasound imaging is the first-line imaging modality to identify the cause of abdominal pain, distension, or mass. It is a helpful way to reveal hydatid sands by shifting the patient's position during the procedure. Also, it reveals the snake/serpent sign when the pressure rises inside the cyst and the inner membrane is detached and the Water-Lily sign when the inner membrane collapses. So, it is an essential tool in evaluating the majority of organs, especially solid organs like the liver. Ct scan is another helpful way which has higher sensitivity of about 95–100%. It helps the physician to determine if the cyst is primary or secondary. In intra-abdominal extra-intestinal cysts, a CT scan is more accurate than ultrasonography. MRI is the most helpful method in the evaluation of soft tissue like the brain. It provides detailed information about the anatomy of the cerebral [2,6]. We cannot depend on laboratory values in diagnosing hydatid cysts if they form in unusual locations and are not complicated. ELISA plays a high role in detection the of cysts in the liver and lungs, but its sensitivity and specificity decrease when the cyst forms in other locations because of the insulating layer which prevents the host's immune response. The liver function test also remains normal when the cyst is inter the abdomen [6]. The treatment methods vary depending on the number and location of the cysts and the patient's condition, including surgery, puncture aspiration injection respiration (PAIR), and chemotherapy. In some cases, we just wait and observe. Surgery remains the cornerstone of treatment, it is divided into radical and conservative approaches. It removes the cyst and all its related tissue and successfully leads to a complete cure. It is the first line of treatment in the simple forms of the disease when we have a limited number of cysts in locations that can be reached by safe operation without endangering the patient's life. We avoid surgery in some situations like patients with several cysts in many organs and patients with a high surgical risk. In such conditions, PAIR or chemotherapy can be considered alternative options for treatment [11,12]. We use chemotherapy with benzimidazoles (albendazole or mebendazole) either in association with surgery or alone if the surgery is not indicated. Chemotherapy plays a role in decreasing the recurrence rate, especially in cases

where the cyst is complicated or forms in rare locations [12]. scolicalid agents also have been used to prevent the spread of infection during operation. Hypertonic saline is the most useful one because it is the least induced immune reaction [6].

4. Conclusion

Our case highlights an important condition that endangers the patient's life if it is not treated quickly and carefully. Also, it is a rare situation and often forgotten. So diagnosing and treating such unique situations are a big challenge, especially in low-income countries, where the equipment is not developed and available all the time. In addition, this case raises many questions about the physiology of hydatid disease and the reasons that lead to this unusual location and findings. And clarifies that hydatid disease is a differential diagnosis of a severe condition of abdominal pain. So, Treatment of such cases requires a lot of caution and accuracy to prevent the disease from spreading in the body.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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