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Hepatic hydatid cyst presenting as a cutaneous fistula

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Abstract:

Hepatic hydatid cysts are usually asymptomatic. Nevertheless, they may rupture, causing anaphylactic shock or fistulation. Cutaneous fistulae caused by ruptured hepatic hydatid cysts are extremely rare. Herein, we report a case of infected cutaneous fistula caused by a ruptured hepatic hydatid cyst. A 57-year-old man presented to Al-Ain Hospital complaining of swelling in his right upper quadrant (RUQ) of 5 months' duration. The abdomen was soft, having a fluctuant tender swelling of 12 cm × 15 cm in the RUQ associated with a pus discharging fistula. The patient was admitted with a provisional diagnosis of abdominal wall abscess with pending sepsis. Surgical incision and drainage were performed under general anesthesia. Initially, around 15 ml of pus was drained, followed by the removal of multiple sized transparent cysts typical of hydatid disease. Postoperative abdominal computed tomography (CT) scan showed multiloculated hepatic cysts in the sixth, seventh, and left lobes with the involvement of the abdominal wall. The patient was treated with oral albendazole 400 mg twice daily for 30 days. Repeated CT scan at 4-month follow-up showed a significant reduction of size of the cysts, indicating proper response to treatment. A cutaneous fistula as a complication of a ruptured hepatic hydatid cyst is extremely rare. Awareness of this complication, especially in endemic areas, and using proper imaging and serological tests are vital for reaching a proper diagnosis.

Keywords:

Complications, cutaneous fistula, hydatid cyst, liver

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Introduction

Hepatic hydatid cysts are usually asymptomatic. Nevertheless, they may rupture, causing anaphylactic shock or fistulation.^[1,2] Cutaneous fistulae caused by ruptured hepatic hydatid cysts are extremely rare.^[1,3] Herein, we report a case of infected cutaneous fistula caused by a ruptured hepatic hydatid cyst.

Case Report

The patient gave his informed written consent to publish his case and clinical images.

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A 57-year-old man presented to Al-Ain Hospital complaining of a swelling in his right upper quadrant (RUQ) of 5 months' duration, which was increasing in size. The patient had a laparotomy performed 2 years ago for swelling of unknown etiology. His temperature was 36.8°C, his pulse was 99 bpm, and his blood pressure was 117/69 mmHg. His abdomen was soft, having a fluctuant tender swelling of 12 cm × 15 cm in the RUQ associated with a pus discharging fistula. C-reactive protein was extremely high (423 mg/l), white blood count was normal (7000 × 10⁹), total bilirubin was raised (30.5 μmol/l), while serum albumin was low (26 g/l). The patient was admitted with a provisional diagnosis of abdominal wall abscess with pending sepsis. Surgical incision and drainage were

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performed under general anesthesia. Initially, around 15 ml of pus was drained, followed by the removal of multiple sized transparent cysts [Figure 1] typical for hydatid cysts [Figure 2].

Postoperative abdominal computed tomography (CT) scan showed multiloculated hepatic cysts in the sixth, seventh, and left lobes with the involvement of the abdominal wall [Figure 3a]. Pus was positive for methicillin-resistant staphylococcus aureus (MRSA). Accordingly, the patient was treated with intravenous vancomycin 1 g twice daily and oral albendazole 400 mg twice daily. The patient was discharged on the 4th day after surgery, with oral albendazole 400 mg twice daily for 30 days and oral doxycycline 100 mg twice daily for 10 days. Repeated CT scan at 4-month follow-up showed a significant reduction of size of the cysts, indicating proper response to treatment [Figure 3b]. We referred our patient to a specialized hepatobiliary center, but he preferred to travel back to his own country to continue his treatment. He was lost to follow-up.

Discussion

Cystic echinococcosis is a zoonotic infection causing >95% of echinococcal diseases in humans.^[3] It is a major



Figure 1: Incision and drainage of a fluctuant tender swelling of 12 cm × 15 cm in the right upper quadrant which was associated with a pus discharging fistula yielded 15 ml of MRSA pus followed by removal of multiple sized transparent cysts

public health problem in sheep-raising countries. The life cycle of this parasite includes both sheep and dogs, with human beings infected as an intermediate host by accidental ingestion of dog feces. Hydatid cysts can be diagnosed as an incidental finding during radiological workup or can be symptomatic. A hydatid cyst may increase up to 5 cm per year or maybe unchanged for many years.^[4]

Hydatid cysts may rupture, both internally and externally.^[5] Internal rupture may occur within the biliary tract, in the gastrointestinal tract, or in the peritoneal cavity, whereas external rupture leads to cutaneous fistulation. Cutaneous fistulation is extremely rare. It occurs in several stages, including protrusion of the cyst through the inner muscular abdominal wall followed by involving the subcutaneous soft tissue and finally rupturing into the skin, forming a fistula as occurred in our patient.^[3] Clinical suspicion of such rare complications in endemic areas is essential for making the proper diagnosis when followed by proper radiological and serological investigations.^[3,4] We did not suspect such diagnosis before surgery simply because hydatid disease is very rare in our region and the clinical appearance mimicked an abscess. Accordingly, we did not perform abdominal ultrasound or CT scan before surgery. Nevertheless, we acknowledge that the previous surgery, the raised bilirubin, and the low albumin should have raised suspicion and prompted a more thorough preoperative diagnostic workup in this patient including abdominal ultrasound and CT scan.

There is no consensus on the management of hepatic hydatid disease. Its management is based upon patient's adherence to long-term monitoring, cyst characteristic, and available medical and surgical experts.^[4,6] Although medical therapy is inadequate, albendazole or in combination with praziquantel is effective as adjuvant



Figure 2: The classical appearance of different sizes of transparent hydatid daughter cysts that were removed through the wound

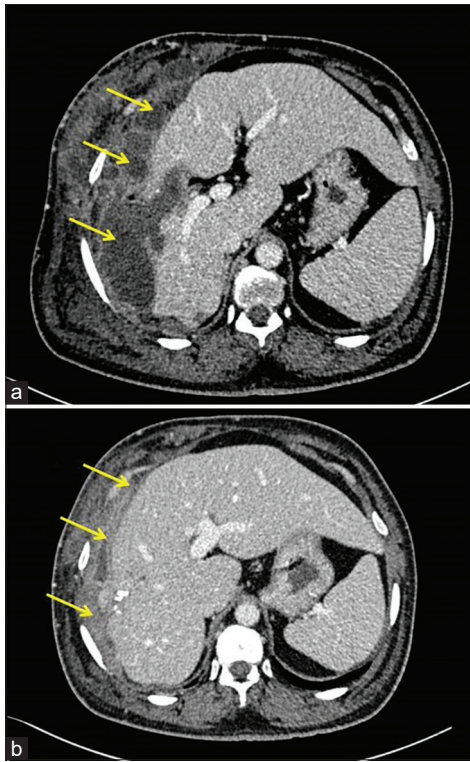


Figure 3: Postoperative abdominal computed tomographic scan with intravenous contrast (a) showing multiloculated hepatic cysts in the sixth and seventh lobes (yellow arrows) and thickened right-sided abdominal wall. Follow-up abdominal computed tomographic scan with intravenous contrast at the same level (b) showing significant regression of the size of the cysts indicating proper response to medical treatment

therapy. The rate of postoperative recurrence can reach up to 10%,^[7] which is less in patients who receive albendazole therapy.^[8] The primary aim of surgery is to remove the parasitic tissues completely.^[4] Surgery varies from cyst unroofing, percutaneous aspiration of cysts, total pericystectomy, subtotal pericystectomy, or hepatectomy.^[9]

Conclusions

A cutaneous fistula as a complication of a ruptured hepatic hydatid cyst is extremely rare. Clinical diagnosis of this pathology is difficult, especially when no cystic material is draining from the fistula. Awareness of the hydatid disease complications, especially in endemic

areas, and using proper imaging and serological tests are vital for reaching a proper diagnosis.

Consent to participate

The patient gave his informed written consent to publish his case and clinical images.

Author contribution statement

MABK: participated in the idea, read the literature, wrote the first draft of the paper, and approved the final version of the paper. MIA: participated in the idea, operated on the patient, took the intraoperative images, interpreted the data, summarized the case report, took the patient consent for publication, and approved the final version of the paper. HM was the treating consultant surgeon, participated in the idea, read and approved the final version of the paper. FAZ: participated in the idea, interpreted the data, supervised the writing process, repeatedly edited the first version, answered the reviewers and approved the final version of the paper.

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Conflicts of interest

There are no conflicts of interest.

References

1. Sayek I, Tirnaksiz MB, Dogan R. Cystic hydatid disease: Current trends in diagnosis and management. *Surg Today* 2004;34:987-96.
2. Jayant K, Agrawal S, Agarwal R, Dayama KG. Spontaneous external fistula: The rarest presentation of hydatid cyst. *BMJ Case Rep* 2014;2014. pii: bcr2014203784.
3. Bahce ZS, Akbulut S, Aday U, Demircan F, Senol A. Cutaneous fistulization of the hydatid disease: A PRISMA-compliant systematic review. *Medicine (Baltimore)* 2016;95:e4889.
4. Brunetti E, Kern P, Vuitton DA; Writing Panel for the WHO-IWGE. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. *Acta Trop* 2010;114:1-6.
5. Salerno S, Cracolici E, Lo Casto A. Subcutaneous rupture of hepatic hydatid cyst: CT findings. *Dig Liver Dis* 2006;38:619-20.
6. Anadol D, Ozçelik U, Kiper N, Göçmen A. Treatment of hydatid disease. *Paediatr Drugs* 2001;3:123-35.
7. Gourgiotis S, Stratopoulos C, Moustafellos P, Dimopoulos N, Papaxoinis G, Vougas V, et al. Surgical techniques and treatment for hepatic hydatid cysts. *Surg Today* 2007;37:389-95.
8. Arif SH, Shams-Ul-Bari, Wani NA, Zargar SA, Wani MA, Tabassum R, et al. Albendazole as an adjuvant to the standard surgical management of hydatid cyst liver. *Int J Surg* 2008;6:448-51.
9. Stoot JH, Jongsma CK, Limantoro I, Terpstra OT, Breslau PJ. More than 25 years of surgical treatment of hydatid cysts in a nonendemic area using the "frozen seal" method. *World J Surg* 2010;34:106-13.