



A case of *Echinococcus granulosus* hepatic hydatid cyst together with pyogenic liver abscess in a Nepali patient

Ranjit Sah¹ · Michele Calatri² · Samikshya Neupane³ · Sagar Poudyal³ · Rafael Toledo⁴ · Lucrecia Acosta⁵

Received: 4 December 2019 / Accepted: 7 March 2020 / Published online: 18 March 2020
© Indian Society for Parasitology 2020

Abstract Hydatidic disease (echinococcosis) is a significant health problem in Nepal. In humans, mainly involves liver and commonly remains silent without symptoms or causing only mild pressure symptomatology. However, inadequate invasive medical procedures may cause the rupture of the cyst with serious complications, anaphylactic shock and death. Herein, we report the case of a Nepali patient who had been treated of a hepatic abscess with percutaneous drainage and antibiotics in a previous hospitalization. Study by drain fluid, ultrasonography and the magnetic resonance imaging revealed the presence of an intact Hydatidic cyst in the liver concomitantly with a percutaneously draining abscess (with free hooklets and a drain tube). This emphasizes the importance of putting the Hydatidic cyst into the differential diagnosis of liver abscess, especially in non-endemic areas.

Keywords *Echinococcus granulosus* · Hepatic hydatid cyst · Liver abscess · Nepal

Introduction

Cystic echinococcosis, caused by *Echinococcus granulosus*, is a globally distributed zoonotic helminthiasis caused by *E. granulosus*. *E. granulosus* adult worm parasitizes the intestine of various species of carnivores. Humans, herbivores and omnivores may act as intermediate host when accidentally ingesting the eggs from a contaminated environment (Devleeschauwer et al. 2014; Cicero et al. 2017). In humans, the developed embryo passes through the intestinal wall to enter the portal system reaching, most frequently, the liver where the hydatid cyst is formed. In an intact cyst, the damage is essentially mechanical, with the possibility of an obstruction of the portal vein or the bile duct (Cicero et al. 2017). Small growing cysts are often asymptomatic and may be unnoticed (Scherer et al. 2009). However, the rupture of the cyst may rise complications such as secondary cysts or allergic reactions or anaphylactic shock (Bhandari and Shahi 2019). In Nepal, hydatid disease is common and, probably, its prevalence is underestimated (Joshi et al. 2004), and the actual incidence should be explored further. Herein, we report a case of a Nepali patient with a hepatic hydatid cyst together with pyogenic liver abscess in a Nepali patient.

Case presentation

A 47 years-old Nepalese man who was admitted to the Institute of Medicine (IOM) in Kathmandu. Before admission, he was treated in Abu Dhabi while he was working there. There, he has been recently diagnosed with diabetes mellitus and hypertension together with a history of fever with chills and rigor for 3 weeks and right upper quadrant pain of abdomen with feeling of abdominal fullness for 1 month. The patient left to United Arab Emirates

✉ Lucrecia Acosta
lacosta@umh.es

¹ Institute of Medicine, Tribhuvan University, Kathmandu, Nepal

² Faculty of Medicine and Surgery, University of Cagliari, Sardinia, Italy

³ Institute of Medicine, Tribhuvan University Teaching Hospital, Kathmandu, Nepal

⁴ Universidad de Valencia, Valencia, Spain

⁵ Área de Parasitología, Departamento de Agroquímica y Medio Ambiente, Universidad Miguel Hernández de Elche, Campus de Sant Joan, 03550 Sant Joan, Alicante, Spain

and returned to Nepal on his own on a commercial flight without medicalized air transfer. When he presenting to us in (IOM), he had percutaneous abdomen draining tube in situ with brownish black fluid in the drainage bag, and persistent fever, anorexia and weight loss of more than 13 kg within 1 month. We treated him with injectable antibiotics and maintained hemodialysis for 2 weeks because of a provisional diagnosis of pyogenic liver abscess and chronic kidney disease in a hospital in Abu Dhabi. A complete blood count revealed leucocyte count was 3840/ μl (58% of neutrophils) and platelet 221,000/ μl and the hemoglobin 8.2 gm/dl. Creatinine was to 630 $\mu\text{mol/L}$. Bilirubin and liver enzymes were within normal limits except raised alkaline phosphatase of 723 IU/L.

He had tender palpable liver with percutaneous drainage in situ and no sign of ascites. Ultrasonography and magnetic resonance imaging were performed to verify its integrity. The magnetic resonance (Fig. 1a, b) and the ultrasonography (Fig. 1c) revealed an intact cyst, with a drain tube outside of cyst in the liver. However, the drainage fluid microscopy examination showed various hooklets with motile bacilli (Fig. 1d). The hooklets were characteristics of *Echinococcus granulosus* and the serology for *E. granulosus* was positive by ELISA in Tribhuvan University Teaching Hospital (Kathmandu, Nepal). The

culture of drainage fluid grew *Pseudomonas* and *Enterobacter* on aerobic culture and the treatment was started immediately after diagnosis. In addition, the photographs of drainage fluid were sent to the CDC (Atlanta, USA) and they confirmed as Hydatidic sand of *Echinococcus* species.

After treatment with albendazol and antibiotics, surgery was performed. The size was on 14th June 5.7 \times 7.2 cm and on 15th June 8.6 \times 7.6 cm of cyst was noted. Histopathological examination of the extracted cyst confirmed the diagnosis of *E. granulosus* (Fig. 2). The removal of the whole cyst resulted the recovery and discharge of the patient.

Discussion

Most relevant features of our case were the presence of an intact cyst together with free hooklets and concomitant infection with several Gram negative bacilli. Common unspecific symptoms of hydatid disease like right upper quadrant pain with feeling of abdominal fullness were observed. Moreover, we found free hooklets in the drained fluid (Hydatidic sand) of the patient suggesting the existence of an additional cyst previously broken and overinfected when it was drained. The presence of an additional

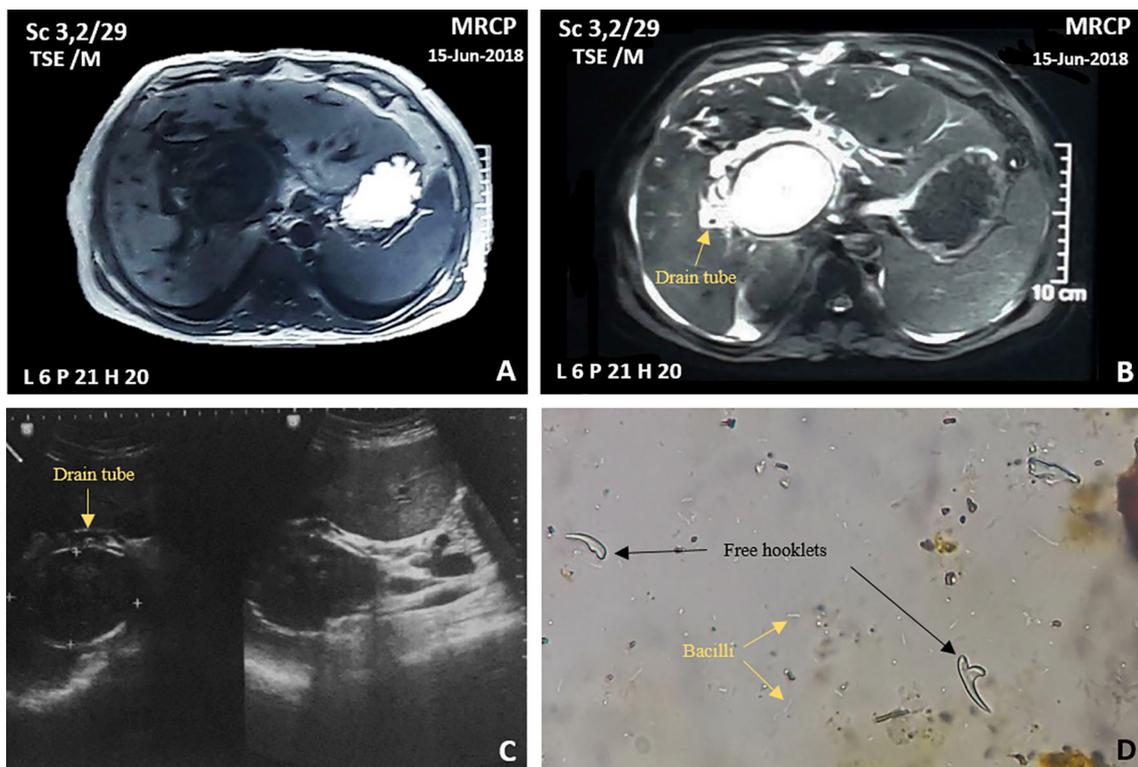
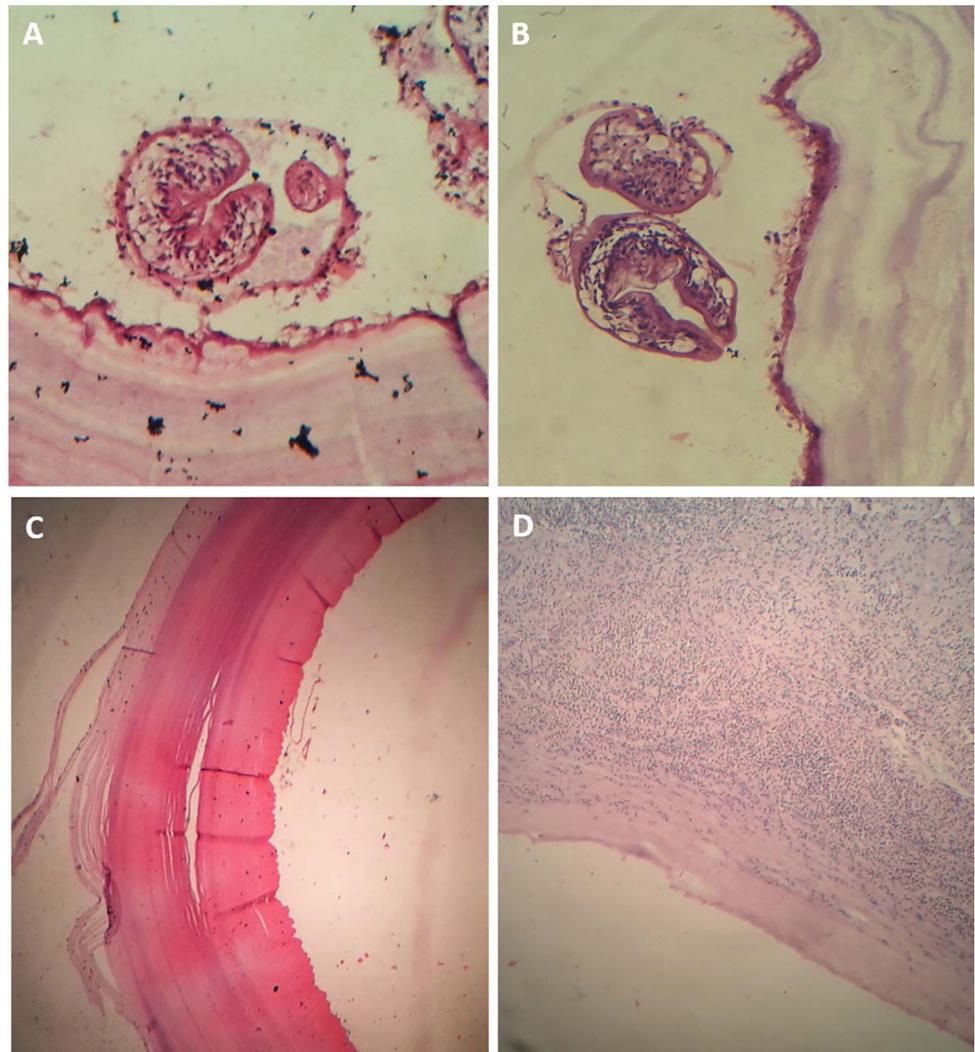


Fig. 1 **a** Magnetic resonance image showing the intact hydatid cyst; **b** magnetic resonance image showing the drain tube outside of intact cyst in the liver abscess **c** ultrasonography revealing the intact hydatid

cyst, with the drain tube outside of intact cyst in the liver abscess drained percutaneously; **d** hepatic drainage fluid showing free hooklets of *Echinococcus granulosus* and gram negative bacilli

Fig. 2 Histopathological examination of the hydatid cyst: **a, b** images showing the protoscolices, brood capsule, germinal layer and laminated membrane of the cyst; **c** detail of the germinal layer and the laminated membrane of the cyst; and **d** fibrous tissue adjacent to the cyst, showing marked inflammation



cyst is supported by the finding of a drain tube by abdominal ultrasonography outside the intact cyst detected by us. The rupture of a cyst into the abdominal cavity constitutes a serious complication (Bhandari and Shahi 2019). Moreover, the rupture of cyst may lead to intraperitoneal dissemination of the protoscolices developing secondary cysts (Cicero et al. 2017; Eckert and Deplazes 2004). The leak of the hydatid fluid and sand can cause an immune reaction to the antigens that may lead to death. In our patient, the allergic and anaphylactic reactions were moderate or absent despite of the signs of a previous percutaneous drainage. However, the lack of marked allergic response could be explained by the by the diabetes of the patient and his immune-compromised condition that could have prevented for the anaphylactic reaction as a result of the cyst rupture. These facts led us not to reject the hypothesis of the existence of an additional cyst previously broken. Our results suggest that our patient could be put at

serious risk to suffer the consequences of the puncture of a hydatid cyst.

Cystic hydatidosis (CH) in Nepal has been mainly studied in animals (Manandhar et al. 2006; Joshi et al. 1997). Devleeschauwer et al. (2014) qualified CH as a probably endemic zoonotic disease in Nepal. Human CH in Nepal was firstly investigated in Kathmandu, where 1194 cases were reported (Joshi et al. 1997). Using coproantigen ELISA test, a prevalence of 11–12% was detected in hospitals and a blood bank from Kathmandu Valley and Banepa (Joshi et al. 1997). Hydatid cyst primarily affects the liver but extrahepatic cysts are common (Reuter et al. 2000). The common extrahepatic sites are lungs, and rarely heart, brain, kidney, bones, pancreas, adrenal or muscles (Meyer and Adam 1989). In Nepal, various authors have reported the presence of a hydatid cyst in unusual presentations: paraspinal muscle (Rauniyar et al. 2012), infratemporal (Thapa et al. 2018), or iliac fossa (Bhandari and Shahi 2019). In our patient, no extrahepatic cyst was

detected and as an intact cyst was detected, surgical intervention and drainage appeared adequate and successfully applied. This emphasizes the need of an accurate and rapid diagnosis of the CH, especially in areas, such as Nepal, where echinococcosis is a common infection (Joshi et al. 1997) such the liver is the anatomical site where a hydatid cyst may become easily infected by various Gram negative microorganisms (Mavilia et al. 2016).

Conclusion

Although the difficulty entailed in the diagnosis of this disease, the determination of the nature of a hepatic abscess may often be challenging, but the identification of the microorganism associated with the infection of the cyst, represents the first step toward the planning of a focused antibiotic therapy and assumes a critical role, nowadays. This case stresses the importance of putting the hydatid cyst into the differential diagnosis together with all the other causes of liver abscess, especially in endemic countries.

Acknowledgements Thanks are given to Dr. Shusila Khadka, Dr. Mohan Khadka, Dr. Rabin Hamal, Dr. Uma Bhatta, Dr. Sanjit Sah, Dr. Ranjana Sah and Dr. Shyam Sah for their role in the management of patient.

Authors' contribution RS, SN and SP made the diagnosis and reviewed the literature. RS and MC analysed the data and contributed to write the manuscript. Both RT and LA analysed the data, drafted the manuscript and designed the figures. LA prepared the manuscript for submission. All authors read and edited the first draft for intellectual content.

Funding The author(s) received no financial support for the research, authorship, and/or publication of this article.

Compliance with ethical standards

Conflict of interest The authors declare that no conflict of interest exists.

Informed consent Written informed consent from the patient was obtained.

References

- Bhandari TR, Shahi S (2019) Simultaneous hydatid cyst of the liver and left iliac fossa: an unusual case report. *Case Rep Surg*. <https://doi.org/10.1155/2019/9101425>
- Cicero G, Blandino A, Ascenti G et al (2017) Superinfection of a dead hepatic echinococcal cyst with a cutaneous fistulization. *Case Rep Radiol*. <https://doi.org/10.1155/2017/9393462>
- Devleeschauwer B, Ale A, Torgerson P et al (2014) The burden of parasitic zoonoses in Nepal: a systematic review. *PLoS Negl Trop Dis* 8(1):e2634. <https://doi.org/10.1371/journal.pntd.0002634>
- Eckert J, Deplazes P (2004) Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. *Clin Microbiol Rev* 17(1):107–135. <https://doi.org/10.1128/CMR.17.1.107-135.2004>
- Joshi DD, Joshi AB, Joshi H (1997) Epidemiology of Echinococcosis in Nepal. *Southeast Asian J Trop Med* 28(S1):26–31
- Joshi DD, Maharjan M, Johansen MV et al (2004) Taeniasis/cysticercosis situation in Nepal. *Southeast Asian J Trop Med Public Health* 35:252–258
- Manandhar S, Hörchner F, Morakote N et al (2006) Occurrence of hydatidosis in slaughter buffaloes (*Bos bubalis*) and helminths in stray dogs in Kathmandu Valley, Nepal. *Berl Munch Tierarztl Wochenschr* 119(7–8):308–311
- Mavilia MG, Molina M, Wu GY (2016) The evolving nature of hepatic abscess: a review. *J Clin Transl Hepatol* 4(2):158–168. <https://doi.org/10.14218/JCTH.2016.00004>
- Meyer E, Adam T (1989) Unusual manifestations of echinococcosis. *Radiologie* 29(5):245–249
- Rauniyar RK, Sharma U, Baboo S (2012) Isolated extra hepatic hydatid cyst of para spinal muscle—unusual presentation—a case report. *NJR* 2(1):31–34
- Reuter S, Seitz HM, Kern P et al (2000) Extrahepatic alveolar echinococcosis without liver involvement: a rare manifestation. *Infection* 28(3):187–192
- Scherer K, Gupta N, Caine WP et al (2009) Differential diagnosis and management of a recurrent hepatic cyst: a case report and review of literature. *J Gen Intern Med* 24(10):1161–1165. <https://doi.org/10.1007/s11606-009-1062-1>
- Thapa S, Ghosh A, Ghartimagar D, Shrestha S et al (2018) Hydatidosis of infratemporal fossa with proptosis—an unusual presentation: a case report and review of the literature. *J Med Case Rep* 12:309. <https://doi.org/10.1186/s13256-018-1812-y>

Publisher's Note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.