CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 73 (2020) 199–202

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Hepato-pericardial fistula revealed by a massive pericardial effusion: A case report of an exceptional complication of the hydatid liver cyst

Ayoub Abetti[a,b,*], Selma Lyazidi[a], Zakaria Qechchar[c], Rachida Habbal[c], Youssef Ettaoumi[a]

[a] Department of Cardiovascular Surgery, Ibn Rochd University Hospital, Casablanca, Morocco
[b] Department of Cardiovascular Surgery, Mohammed VI University Hospital, Faculty of Medicine and Pharmacy, Oujda, Morocco
[c] Department of Cardiology, Ibn Rochd University Hospital, Casablanca, Morocco

ARTICLE INFO

Article history:
Received 5 May 2020
Received in revised form 9 July 2020
Accepted 10 July 2020
Available online 16 July 2020

Keywords:
Hydatid cyst
Fistula
Liver
Pericardial effusion
Case report

ABSTRACT

INTRODUCTION: The rupture of the hydatid liver cyst in the pericardium is a very exceptional and serious complication that can cause sudden death following cardiac tamponade or anaphylactic shock.
CASE PRESENTATION: We report a case of a 25 years-old woman with a massive pericardial effusion due to fistulization of hepatic hydatid cyst. Surgical closure of the fistula and the resection of the tow hydatid cysts were successful in managing this rare case.
DISCUSSION: Hepato-pericardial fistula is an extremely rare complication of hydatid liver cyst. Only 6 similar cases were previously reported in the literature. The Hepato-pericardial fistula may result in an acute pericarditis that progress to either cardiac tamponade or constrictive pericarditis. Its diagnosis is based on ultrasound and CT imaging. The surgical treatment with supportive therapy seems to improve the outcomes.
CONCLUSION: Pericardial effusion secondary to rupture of hepatic hydatid cyst should always be suspected in endemic countries.

1. Introduction

Hydatidosis or cystic echinococcosis is a parasitic infection associated with the larval form of the dog tapeworm *Echinococcus granulosus* [1]. This parasitosis is a major public health problem in Morocco, as well as the rest of the Maghreb countries. The liver is the most frequently affected organ [1], while pericardial involvement is extremely uncommon entity even in endemic countries [2]. In this paper, we report a very rare case of a large pericardial effusion due to the rupture of the hepatic hydatid cyst into the pericardium.The management of this case was successfully conducted at our university hospital. This work is reported in line with the SCARE 2018 criteria [3].

2. Case report

A 25 years-old female from Western Morocco resident in a rural area, she reported a prolonged contact with dogs. Moreover, the patient has no history of any drugs use, nor family or genetic and other antecedents. She was admitted to the emergency department for the management of retrosternal chest pain associated with worsening exertional dyspnea that was progressing for 12 days. On clinical examination, the patient was apyretic, the cardiac frequency was 86 beats per minute (bpm), the respiratory frequency was 22 cycles per minute (cpm) and the blood pressure was 98/60 mmHg. Cardiovascular examination showed signs of right heart failure with edema of the lower limbs and spontaneous jugular vein dilatation. Moreover, a decrease in heart sounds on auscultation was marked. The abdomino-pelvic examination revealed sensitivity of the right hypochondrium. The electrocardiogram (ECG) showed a regular sinus rhythm at 86 cpm. Chest X-ray revealed the presence of cardiomegaly with elevation of the right diaphragmatic dome. In addition, transthoracic echocardiography (TTE) showed a large circumferential pericardial effusion with intrapericardial vesicles (Fig. 1) and a hepato-pericardial fistula appearance. The thoracoabdominal CT scan revealed the presence of two multivesicular hydatid cysts (type III) affecting segments II and IV of the liver. The hydatid cyst of segment II communicates with the pericardium through a thin aperture causing a large pericardial effusion (Fig. 2). The hydatid serology was positive and the rest of the laboratory tests were normal. Therefore, the diagnosis of pericardial effusion secondary to the hydatid cyst rupture was confirmed and the treatment by oral albendazole (15 mg/kg) was started. Considering the high risk of tamponade and anaphylactic shock, the combined surgical procedures were performed by

* Corresponding author at: Department of Cardiovascular Surgery, Mohammed VI University Hospital, BP 4806 University, 60049, Oujda, Morocco.
E-mail address: abettiayoub06@gmail.com (A. Abetti).

https://doi.org/10.1016/j.ijscr.2020.07.036

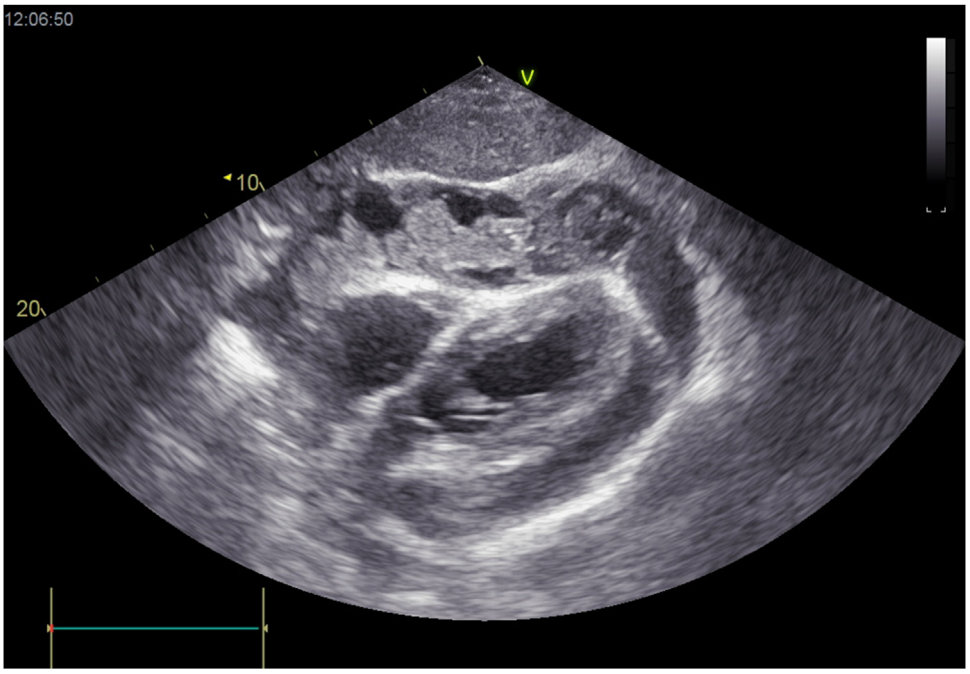

Fig. 1. Echographic image in the subxiphoid view showing a large pericardial effusion containing daughter vesicles.

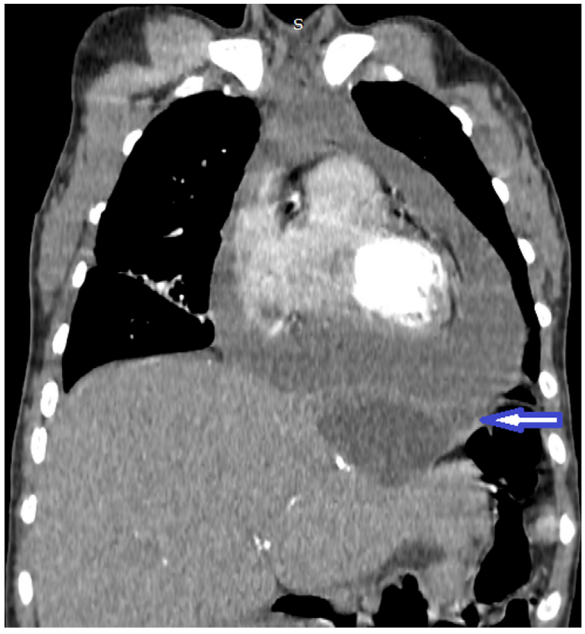

Fig. 2. Thoracic CT angiography in coronal section, showing the hydatid cyst of segment II communicating with the pericardium through a thin aperture causing a large pericardial effusion.

cardiovascular and hepto-biliary Professors assisted by final year cardiovascular surgery residents (3 days after the hospitalization). The first surgery was performed by median sternotomy with longitudinal opening of the pericardium. The aspiration of the pericardial fluid containing the daughter cysts was performed. The perioperative assessment revealed a thickened pericardium (Fig. 3) and the daughter vesicles were evident at the fistula in the left diaphragmatic side of the pericardium (Fig. 4). After abundant washing with hypertonic saline, the fistula was closed with a continuous suture technique using 1 polyglactin 910. The second operative step was performed by midline laparotomy. The procedure consisted of a resection of the hydatid cyst of segment IV of the liver with the protruding dome of the segment II of the liver (Fig. 5). A reinforcement of the closure of the cysto-pericardial fistula by an x-shaped interrupted suture technique was also performed. The patient spent a day in the intensive care unit and six days in the cardiovascular surgery department. She was extubated two hours after surgery and discharged after seven days later. The patient was kept under oral albendazole for the following three months with good adherence and tolerability. The anatomopathological examination of the membranes confirmed the diagnosis of hydatid disease. However, the patient was asymptomatic with no signs of recurrence on echocardiography at the third month of follow up.

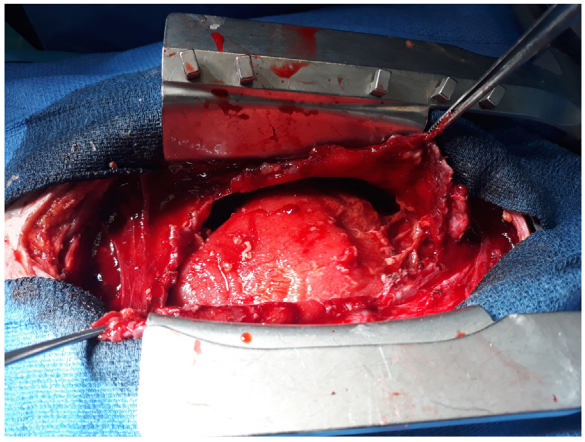

Fig. 3. Perioperative view showing the thickened pericardium.

3. Discussion

Hydatid disease or cystic hydatidosis is a highly-responsive zoonosis in cattle-rearing areas where there is a close contact between humans, dogs and sheep [2]. The highest prevalence is found in the countries of the Mediterranean and Middle East, South America, Africa, Australia and New Zealand [2,4]. Contamination of

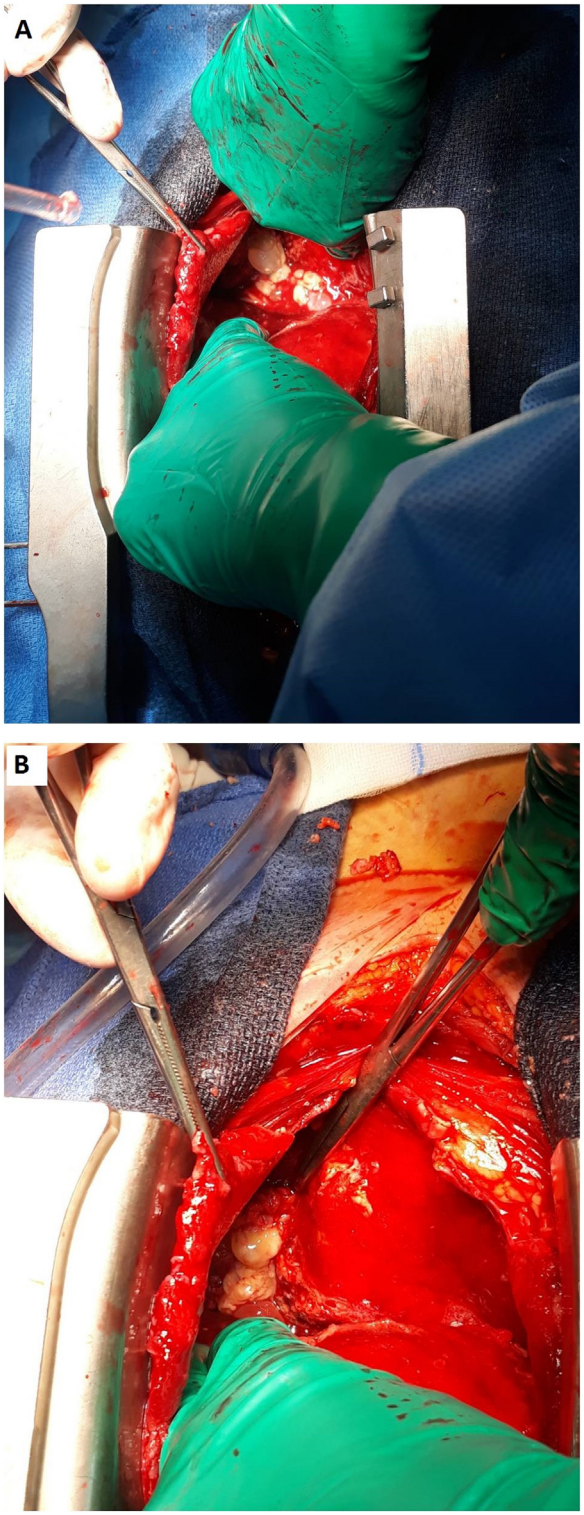

Fig. 4. (A, B): Perioperative view showing the exit of the daughter vesicles through the transdiaphragmatic fistula.

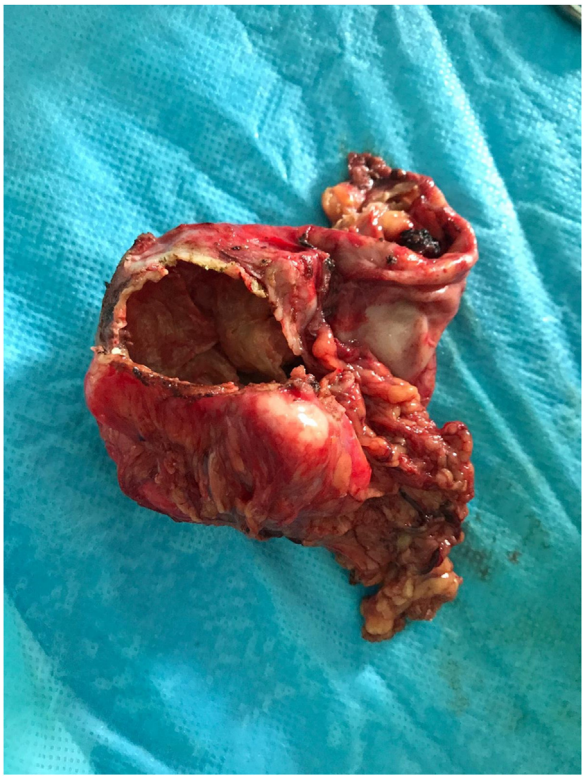

Fig. 5. An image of the liver hydatid cyst after surgical resection.

humans occurs either indirectly by ingesting water or food that are contaminated by the parasite's eggs or directly through contact with infected dogs [1,4]. The hydatid disease can affect all organs of the body, from the head to the feet through the general or lymphatic circulation [2]. The liver is the first line of protection, and is therefore the most frequently affected organ (50%–70%) [1,4]. Cardiac hydatidosis is rare even in highly endemic countries. It occurs in only 0.5–2% of hydatidosis cases [5]. However, pericardial localization without cardiac involvement is extremely rare and always appears to be secondary [5]. In our case, the pericardial involvement was secondary to a rupture of a hydatid cyst of the segment II of the liver in the pericardium through a diaphragmatic fistula. To the best of our knowledge, there are only 6 similar cases reported in the literature of the rupture of hydatid liver cyst in the pericardium until to date [6–11]. Four out of those six cases were located in left hepatic lobe and only 2 cases in the right lobe of the liver. In our case, the cyst was located in the left hepatic lobe. However, all of these cases had a type III of hydatid cyst.

The mechanism of the rupture of the hydatid cysts of the liver in the pericardium can be explained by a phenomenon of friction against the diaphragmatic wall, which leads to the development of adhesions between the cyst and the diaphragm. Therefore, this weakens the cyst wall which causes fistula occurrence [12]. The clinical manifestations are non-specific and diverse, depending on the location, the size, the number and the content of the hydatid cyst. As it develops, the liver hydatid cysts can constrict or rupture in the surrounding tissues [13]. The rupture of the cyst in the pericardial cavity may result in an acute effusion with a pattern of acute serofibrinous or purulent pericarditis that progresses to either cardiac tamponade or constrictive pericarditis [5,12]. In this case, the delay in diagnosis or management may cause death from anaphylactic shock or tamponade [14]. Biologically, the hydatid serology is positive only in about 50% of cases of cardiac hydatidosis [5,15]. Our patient's hydatid serology was positive. Chest X-ray is not very helpful. It shows cardiomegaly with elevation of the right diaphragmatic dome [6,9]. TTE is preferred for the diagnosis of cardiopericardial hydatid cysts. The presence of a membrane detachment, multivesicular patterns or a daughter vesicle in the pericardial cavity is strongly suggestive of hydatid origin [15]. The CT scan confirms the diagnosis and shows the topography of the cyst with the surrounding organs [16]. Importantly, magnetic resonance imaging (MRI) provides a better view of the cysts topography and their anatomical features [17]. Despite therapeutic advances,

surgery is by far the best treatment for complicated hydatid cysts [13]. Our attitude was to treat the patient using albendazole at a dose of 15 mg/kg before and after surgery. Several authors have recommended this supportive therapy as it reduces the risk of spread and recurrence [18,19]. The patient was satisfied by our medical and surgical management.

We noticed that all cases of ruptured hydatid cysts in the pericardium were type III cysts and the majority of them were located in the left lobe of the liver (5 cases). Therefore, we argue that the hydatid cysts with high risk of rupture in the pericardium are type III of the left hepatic lobe. However further research is needed to explain this hypothesis.

4. Conclusion

Pericardial effusion secondary to rupture of hepatic hydatid cyst should always be suspected in endemic countries. Furthermore, the hydatid cyst in the left lobe of the liver especially type III should be closely followed and treated early.

Declaration of Competing Interest

The authors report no conflicts of interest.

Sources of funding

None.

Ethical approval

Not required for this case report.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Dr Ayoub Abetti: writing the manuscript, Review and Editing, Visualisation. Dr Zakaria Qechchar: Conceptualization and Investigation. Professor Selma Lyazidi provided the imaging data of the patient. Professors Rachida Habbal and Youssef Ettaoumi supervised the writing of manuscript.

Registration of research studies

Not required.

Guarantor

Dr. Ayoub Abetti (MD).
Pr. Youssef Ettaoumi (MD).

Provenance and peer review

Not commissioned, externally peer-reviewed.

Acknowledgements

The authors would like to express their gratitude to Dr Fatima Zahra Talhi, Dr Issam Rahal, Dr Saousan Serbout and Dr Adam Belaarbi, for their participation of the management of our patient.

References

[1] R. Blanton, Echinococcosis, in: R.E. Behrman, R.M. Kliegman, H.B. Jenson (Eds.), Nelson Textbook of Pediatrics, 17th edn, WB Saunders Company, Philadelphia, 2004, pp. 1173–1174.
[2] P. Polat, M. Kantarci, F. Alper, S. Suma, M.B. Koruyucu, A. Okur, Hydatid diseasefrom head to toe, Radiographics 23 (2003) 475–494.
[3] R.A. Agha, M.R. Borrelli, R. Farwana, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus Surgical Case Report (SCARE) guidelines, Int. J. Surg. 60 (2018) 132–136.
[4] I. Pedrosa, A. Saiz, J. Arrazola, J. Ferreiros, C.S. Pedrosa, Hydatid disease: radiologic and pathologic features and complications, Radiographics 20 (2000) 795–817.
[5] M. Chellaoui, R. Bouhouch, S. Akjouj, L. Chat, F. Achaabane, D. Alami, A. Najid, H. Benamour-ammar, Hydatidose péricardique : à propos de 3 observations, J. Radiol. 84 (3) (2003), 329–3.
[6] S.R. Ahuja, S. Karande, S.R. Koteyar, M.V. Kulkarni, Hepatic hydatid cyst rupturing into sub-diaphragmatic space and pericardial cavity, J. Postgrad. Med. 47 (2001) 37–39.
[7] F. Martin-Herrero, I. Cruz, L. Muñoz-Bellvis, Hepatic hydatid cyst rupturing into pericardial cavity, Heart (2006), http://dx.doi.org/10.1136/hrt.2006.090456, Article *in* Heart (British Cardiac Society) November.
[8] Ali Can Hatemi, Oğuz Omay, Ulaş Kumbasar, Mustafa Canikoğlu, Kaya Süzer, Constrictive pericarditis due to hepatopericardial fistulas of hepatic echinococcosis, Turk Gogus Kalp Dama 21 (1) (2013), 219 221.
[9] L. Chemaou El Fihri, A. Hadi, D. Benzeroual, M. Elhattaoui, M. El Merdouli, M. El Jamili, Z. Zouizra, D. Boumzebra, R. Abbas, M. Boukhfaoui, Kh Rabbani, Pericardial effusion reveals an hepatopericardial fistulas of hepatic echinococcosis (A case report), Cardiovasc. Disord. Med. (2019), http://dx.doi.org/10.15761/CDM.1000196.
[10] Ozgur Yagmur, Orhan Demircan, Erol Atilla, Ali Alparslan Mustafa Demirtas, Cardiac tamponade due to rupture of a hepatic hy-datid cyst into the pericardium, Dig. Surg. 9 (1992) 329–331.
[11] Sercan ÖZKAÇMAZ, Computed tomography findings of ruptured hepatic hydatid cyst into the pericardial space: a case report, Iran. J. Parasitol. 14 (Oct–Dec (4)) (2019) 674–678.
[12] F. Klotz, X. Nicolas, J.M. Debonne, J.F. Garcia, J.M. Andreu, Kystes hydatiques du foie. EncyclMédChir (Editions Scientifiques et Médicales Elsevier SAS, Paris, tous droits réservés), Hpatologie (2000) 16, 7-023-A-10.
[13] M. Milicevic, Hydatid disease, in: L.H. Blugmart (Ed.), Surgery of the Liver and Biliary Tract, 2nd ed., Churchill Livingstone, Edinburgh, 1994, pp. 1121–1150.
[14] M. Kosecik, M. Karaoglanoglu, B. Yamak, Pericardial hydatidcyst presenting with cardiac tamponade, Can. J. Cardiol. 22 (2) (2006) 145–147.
[15] S. Elkarimi, et al., Tamponnade révélant un kyste hydatique intra-péricardique - un cas, Ann. Cardiol. Angeiol. (Paris) (2014).
[16] J.A. Trigano, F. Mourot, T. Talmoudi, C. Malmejac, J. Torresani, J. Houel, Sémiologie du kyste hydatique du coeur. Étude d'une série continue de 13 cas et intérêt du scanner, Arch. Mal. Coeur. 78 (13) (1985) 1895–1899.
[17] S.E. Golzari, M. SokoutiPericyst, The outermost layer of hydatid cyst, World J. Gastroenterol. 20 (February (5)) (2014) 1377–1378.
[18] C. Dziri, K. Haouet, A. Fingerhut, A. Zaouche, Management of cystic echinococcosis complications and dissemination: where is the evidence? World J. Surg. 33 (Jun. (6)) (2009) 1266–1273.
[19] Shams-Ul-Bari, S.H. Arif, A.A. Malik, A.R. Khaja, T.A. Dass, Z.A. Naikoo, Role of albendazole in the management of hydatid cyst liver, Saudi J. Gastroenterol. 17 (Sep–Oct (5)) (2011) 343–347.