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Case Report

Primary splenic hydatid cyst an unexpected diagnosis: Case report

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ABSTRACT

Introduction: Hydatid disease is endemic in farming areas but occurs worldwide. The most common site of disease is liver. Hydatid disease of the spleen is a rare condition. Worldwide incidence of splenic hydatid is 0.5–4%. Surgery is the mainstay of treatment. The standard treatment is open total or partial splenectomy.

Presentation of case: A 28 year-old female patient was referred to our institution, after coincidental sonography finding. On abdominal examination, there was no sensitization, and there was no resistance or rebound. Ultrasound showed an enlarge spleen; an abdominal CT confirmed the presence of a splenic cyst at the lower pole of the spleen of 7 cm in diameter, no other organ involvement. The biological confirmation was made by indirect hemagglutination. Spleen-sparing surgery was performed. Macroscopic and microscopic examination of the specimen confirmed Hydatid cyst. The patient was discharged from hospital on the seventh postoperative day with a prescription for albendazole (2 × 400 mg/day) for three months.

Discussion and conclusion: The rarity of primary splenic hydatid disease poses a diagnostic challenge for clinicians, the disease should be considered in differential in every patient in endemic areas with cystic lesion of spleen until proved otherwise, it may be detected incidentally or present with non-specific complaints, preservation surgery should always be tried to avoid post splenectomy infection, especially in young patients.

1. Introduction

Hydatid Disease (HD) is a zoonosis caused mainly by the larval stage of the cestode worm *Echinococcus granulosus* or dog tapeworm. It is endemic in cattle-rearing areas of South America, Africa, Middle East, South Europe, India, and Australia. HD of the spleen is a rare entity, worldwide incidence of splenic hydatid is 0.5–4% [1].

Parasitic cysts of the spleen are almost exclusively hydatid cysts. In endemic areas, 50–80% of splenic cysts are echinococcal [2].

Few days after ingestion of hydatid eggs, a fluid-filled cyst begins to develop, with subsequent development of multiple layers to become a metacestode (hydatid cyst) which grows at a rate of 0.3 cm–1 cm/year [3,4].

It may be detected incidentally or present with non-specific complaints [1].

The first clinical indication of the presence of splenic hydatid disease is usually an accidentally discovered mass in the abdomen mostly in left hypochondrium [5,6], and less frequently in epigastrium. Pain, usually a dull dragging ache, is often the first clinical sign. There can be dyspepsia, constipation due to pressure on colon, dyspnea due to pushing up of the left diaphragm [7].

Spontaneous rupture of isolated splenic hydatid cyst is reported [8], sometimes hydatid cyst can present as a simple cyst without having the classic serological and imaging features, and later can lead to life-threatening complications like anaphylaxis [9], calcification of the cyst may also occur.

Owing to the risk of spontaneous or traumatic rupture, splenic hydatid cysts are usually treated surgically. Spleen-preserving surgery like

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partial splenectomy is a risk factor for recurrence and post-operative haemorrhage, but on the other hand, total splenectomy predisposes to sepsis and this should be avoided especially in children.

The aim of this case report is to emphasize that preservation of spleen should always be tried to avoid post splenectomy infection, and mortality in the postoperative period. This work has been reported in line with the SCARE criteria [10].

2. Presentation of case

A 28 year-old female patient was referred to our general surgery department, after coincidental sonography finding, there was no history of pet dogs or sheep at home.

On abdominal examination, there was sensitization, and there was no resistance or rebound. Her mean arterial pressure was 110/750 mmHg, routine laboratory tests did not show any abnormalities.

CT abdomen confirmed the USG findings demonstrating evidence of well defined complex cystic lesion of approximate size of 7 × 5cm at the lower pole of the splenic parenchyma with no evidence of calcification, suggestive of hydatid cysts (Fig. 1). There was no involvement of liver or other organs.

Patient was kept on albendazole 400 mg oral tablet twice per day for 3 weeks before the date of surgery. An Indirect hemagglutination (HAI) was positive for *Echinococcus* sp (titer>160).

Laparotomy was performed through a midline incision. Surgical exploration revealed a hydatid cyst occupying the lower pole of the spleen. Spleen-preserving surgery was performed (Fig. 2), the abdomen was washed locally with hypertonic saline solution (NaCl 20%). Macroscopic and microscopic examination of the specimen confirmed Hydatid cyst.

The patient was discharged from hospital on the seventh post-operative day with a prescription for albendazole (2 × 400 mg/day) for three months, and postoperative recovery was uneventful, no local recurrence was detected during postoperative follow up.

3. Discussion

Hydatid cyst is endemic in cattle-rearing areas of Africa. Almost any anatomic location can be the host site of the parasitic cysts [11,12]. However, isolated splenic hydatid cyst is a very rare manifestation [8]. Worldwide incidence of splenic hydatid is 0.5–4% [1].

Hydatidosis should be suspected in patients with splenic cystic lesions, particularly in endemic areas until proved otherwise [13].

Hydatid cyst of spleen seems to be a coincidental finding [14], and up to 30% of the cases are in asymptomatic individuals [15,16], which is consistent with our observation. The hydatid cysts grow to 5–10 cm in size within the first year and can be asymptomatic for years or even

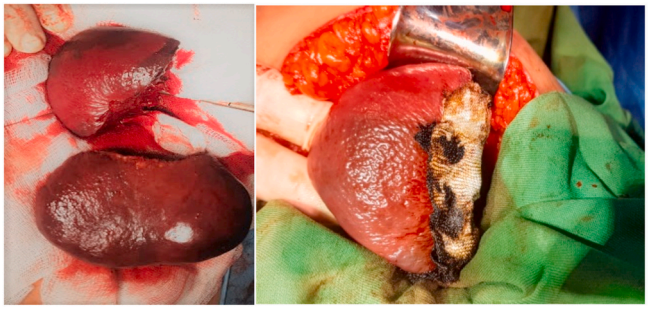

Fig. 2. Per-operatory spleen partial resection.

decades [11], when the cyst reaches considerable size patients may develop symptoms like abdominal pain, enlarged spleen, fever, and a palpable mass.

Hematemesis as a presenting sign has been described in literature [14]. There can be dyspepsia, constipation due to pressure on colon, dyspnea due to pushing up of the left diaphragm, fistula of the colon or perforation into the diaphragm or bronchial tree [6,7], the clinical picture is dramatic in cases with spontaneous rupture into the free peritoneal space which may cause fatal massive hemoperitoneum and anaphylactic reactions. Case reports of acute abdomen caused by spontaneous rupture into abdominal cavity are requiring splenectomy [17].

The differential diagnosis of such lesions includes epidermoid and dermoid cysts, large solitary abscess or hematoma, cystic hemangiomas, intrasplenic pancreatic pseudocyst, and cystic neoplasm of the spleen (lymphangiomas), true cysts are very rare [16,18].

The rarity of splenic hydatid disease poses a diagnostic challenge for clinicians [9]. Rarely, patients present late after the cyst is ruptured [8]. In the asymptomatic patients, the diagnosis is established during investigations for other reasons, or when the cyst reaches an advanced size [16].

Imaging combined with immunological tests solves the diagnostic problem most of the times [9]. Radiological findings are similar to those of liver hydatid cysts, ranging from cystic to solid appearance and from a unilocular cyst to multivesicular cyst. Calcifications in the cyst wall may occur. The classic findings on imaging of hydatid cysts are double layer thick cyst walls, often with daughter cysts [11]. CT facilitates diagnostic and therapeutic procedures in order to ensure complete surgical resection [9]. The main problem in the diagnosis of splenic hydatidosis is in differentiating it from other splenic cystic lesions that have similar appearances on sonography and computed tomography (CT) [15].

Indirect hemagglutination (HAI), enzyme-linked immunosorbent assay (ELISA), or Western blott are carried out for diagnosis, screening and postoperative follow up for recurrence; but these tests are often

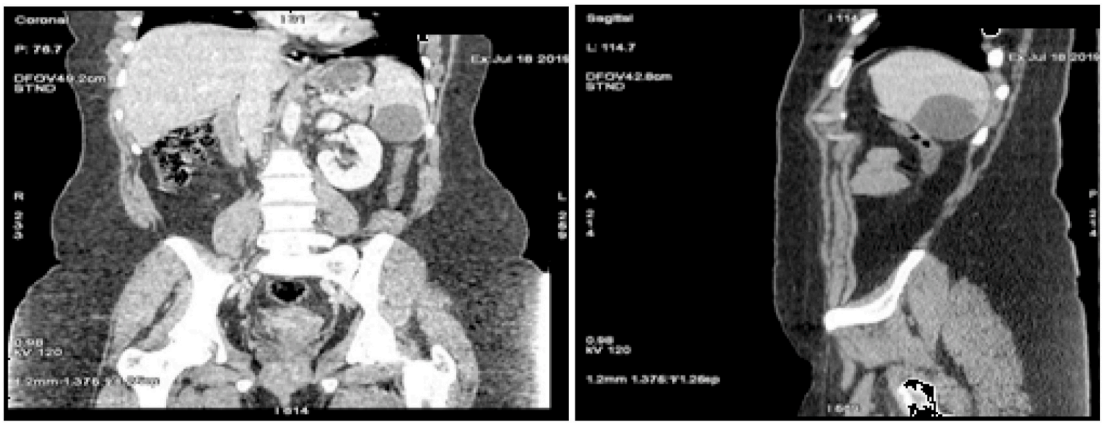

Fig. 1. CT appearance of a primary hydatid cystat the lower pole of the spleen.

negative or inadequate for definitive diagnosis if the cyst is intact, calcified or sterile [19].

Imaging when combined with serological tests like ELISA, HAI and immune-electrophoresis can lead to a successful diagnosis of splenic hydatid cyst in 90% of cases [20].

Splenectomy is the emergency operative management [8], spontaneous rupture into abdominal cavity is requiring total splenectomy [17]. The standard treatment is total or partial splenectomy. Cyst fluid can be drained with puncture and aspiration to reduce the intracystic pressure, but splenectomy without puncturing the cyst is preferable [2].

Spleen-preserving surgery like parietal splenectomy, cyst enucleation, deroofing of cyst with omentoplasty or external drainage are in vogue [9]. Partial splenectomy was therefore introduced, initially for trauma and later for most benign splenic diseases, including non-parasitic cysts, and parasitic hydatid cysts [2]. Laparoscopic splenectomy is being increasingly performed at advanced laparoscopic centers. However, some authors believe it is unsafe due to the risk of anaphylactic shock and intra-peritoneal dissemination. Spillage of protoscolex-rich fluid during surgery occurs in 5%–10% of cases [21].

4. Conclusion

HD is common in liver and lung but no structure of body is exempted from involvement of the hydrated cyst. It means the hydrated cyst can occur in any part of the body. As the hydatid cyst can present as a simple cyst without having the classic clinical, serological and imaging features, primary hydatid disease of spleen should be considered in differential in every patient in endemic areas with cystic lesion of spleen until proved otherwise. Surgery is the mainstay in hydatid cyst disease, Spleen-preserving surgery should always be tried to avoid post splenectomy infection, especially in young patient.

Ethical approval

Patient gave consentement for photos and publication, in accordance with ethical committee of our Hospital. Written informed consent was obtained from participant. Consent from legally authorized representatives parents. Personal details of patient in any part of the paper or supplementary materials were removed before submission.

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Author contribution

Study concept and design Yassine Merad, Ahmed Zeggai.
Writing paper Yassine Merad, Hichem Derrar.
Data collection Ahmed Zeggai, Yassine Merad.
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Research registration number

1. Name of the registry:
2. Unique Identifying number or registration ID:
3. Hyperlink to your specific registration (must be publicly accessible and will be checked):

Guarantor

Yassine Merad, I am the principal author and I accept full responsibility for the work and the conduct of the study, and I control the decision to publish with agreement of co-authors.

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.

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Declaration of competing interest

The authors have no conflicts of interest to disclose.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102293.

References

[1] W.G. Humphreys, G.W. Johnston, Splenic cysts: a review of 6 cases, Br. J. Surg. 66 (1979) 407–408.
[2] K.I. Gharaibeh, Laparoscopic excision of splenic hydatid cyst, Postgrad. Med. 77 (2001) 195–196.
[3] M. Safioleas, E. Misiakos, C. Manti, Surgical treatment for splenic hydatidosis, World J. Surg. 21 (1997) 374–377.
[4] M.A. Dar, O.J. Shah, N.A. Wani, F.A. Khan, P. Shah, Surgical management of splenic hydatidosis, Surg. Today 32 (2002) 224–229.
[5] S.I. Schwartz, Principles of Surgery, seventh ed., McGraw-Hill Companies, New York, 1999, pp. 1395–1435. Chapter 31-Spleen.
[6] C. Uriarte, N. Pomares, M. Martin, A. Conde, N. Alonso, M.G. Bueno, Splenic hydatidosis, Am. J. Trop. Med. Hyg. 44 (1991) 420–423.
[7] I. Alfageme, M. Martin, J. Hernandez, C. Huertas, Rupture of a long-standing splenic hydatid cyst into the bronchial tree, Clin. Infect. Dis. 19 (1994) 992–994.
[8] S. Acharya, B. Ghimire, N. Khanal, Spontaneous rupture of isolated splenic hydatid cyst without acute abdomen: a case report, Clin. Case Rep. 7 (11) (2019).
[9] K. Rasheed, S.A. Zargar, A.A. Telwani, Hydatid cyst of spleen: a diagnostic challenge, N. Am. J. Med. Sci. 5 (1) (2013) 10–20.
[10] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, SCARE group, The SCARE 2020 statement: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[11] P. Polat, M. Kantarci, F. Alper, S. Suma, M.B. Koruyucu, A. Okur, Hydatid disease from head to toe, Radiographics 23 (2) (2003) 475–494.
[12] I. Pedrosa, A. Saiz, J. Arrazola, J. Ferreiros, C.S. Pedrosa, Hydatid disease: radiologic and pathologic features and complications, Radiographics 20 (3) (2000) 795–817.
[13] M.C. Goel, M.R. Agarwal, A. Misra, Percutaneous drainage of renal hydatid cyst: early results and follow-up, Br. J. Urol. 75 (1995) 724–728.
[14] I.J. Klompmaker, E.B. Haagsma, M.J. Slooff, Splenic vein obstruction due to a solitary echinococcal splenic cyst, resulting in gastric fundus varices: an unusual cause of variceal bleeding, HPB Surg. 6 (1993) 229–232.
[15] M.A. Qureshi, C.D. Hafner, Clinical manifestations of splenic cysts: study of 75 cases, Am. Surg. 31 (1965) 605–608.
[16] V. Durgun, S. Kapan, M. Kapan, I. Karabiçak, F. Aydogan, E. Goksoy, Primary splenic hydatidosis, Dig. Surg. 20 (2003) 38–41.
[17] N. Herrera Merino, M. Díaz del Río Botas, A. Marinelli Ibarreta, A. San José Borreguero, J. Abascal Morte, A. Colas Vicente, Acute abdomen caused by spontaneous rupture of splenic hydatid cyst, Rev. Esp. Enferm. Dig. 78 (1990) 102–104.
[18] T. Franquet, M. Montes, F.J. Lecumberri, J. Esparza, J.M. Bescos, Hydatid disease of the spleen: imaging findings in nine patients, AJR Am. J. Roentgenol. 154 (1990) 525–528.
[19] I. Wani, R.A. Wani, K.A. Wani, Primary hydatid of psoas muscle, Urol. Ann. 2 (2) (2010) 89.
[20] M.M. Pukar, S.M. Pukar, Giant solitary hydatid cyst of spleen-A case report, Int. J Surg. Case Rep. 4 (4) (2013) 435–437.
[21] D.M. Culafic, M.D. Kerkez, D.D. Mijac, N.S. Lekic, V.I. Rankovic, D.D. Lekic, et al., Spleen cystic echinococcosis: clinical manifestations and treatment, Scand. J. Gastroenterol. 45 (2) (2010) 186–190.