CASE REPORT

A rare presentation of splenic tuberculosis with a pseudocyst

Kalyan Chakradhar,¹ Shiva Prasad,¹ Sampath Kumar,¹ Manna Valiathan²¹Department of General Surgery, Kasturba Medical College, Manipal, Karnataka, India²Department of Pathology, Kasturba Medical College, Manipal, Karnataka, India**Correspondence to**

Dr Kalyan Chakradhar, chakradharkalyan@yahoo.co.in

Accepted 16 April 2014

SUMMARY

Splenic tuberculosis is an unusual manifestation of extrapulmonary tuberculosis, especially in immunocompetent hosts. It often poses diagnostic difficulties as microbiological confirmation is not easy. In this case report, we describe a case of pseudocyst of the spleen due to tuberculosis, which is a rare clinical presentation with only one case having been reported previously.¹ A 24-year-old immunocompetent woman presented with pain in the abdomen, vomiting and fever. On evaluation the patient was found to have a cyst arising from the spleen. Splenectomy was performed. Pathological examination of the specimen revealed a pseudocyst with granulomas and giant cells consistent with tuberculosis. Cyst fluid analysis revealed lymphocyte predominance with high adenosine deaminase levels. The patient was started on 6 months of antituberculous therapy postoperatively. On follow-up, the patient was found to have gained weight and showed signs of well-being.

BACKGROUND

Tuberculosis (TB) is a major health problem in India. TB manifests with diverse clinical symptoms most commonly as pulmonary TB. Splenic TB usually occurs as a part of miliary TB in immunocompromised patients and rarely as an isolated splenic TB in an immunocompetent individual.² Spleen is the third most common organ involved in miliary TB after lung and liver.³ Splenic cysts are classified as true cysts, which can be parasitic or non-parasitic, or as pseudocyst based on the presence or absence of the epithelial lining, respectively.⁴ TB of the spleen with an associated pseudocyst in an immunocompetent individual is an extremely rare presentation. The literature review showed evidence of only one such case reported previously.¹

CASE PRESENTATION

A 24-year-old woman presented with a dull pain in the upper abdomen since the last 3 weeks, with radiation to the left shoulder and non-bilious vomiting following food intake. She also reported loss of appetite and low-grade intermittent fever. The patient noticed fullness in the upper abdomen a month ago, which was gradually increasing in size. There was no history of trauma, alcohol consumption and contact with pets. No history suggestive of TB could be elicited.

On examination the patient was thin built, stood at a height of 152 cm, weighed 38 kg and had a body mass index of 16.4 kg/m². The patient was febrile with a temperature of 39.4°C; pallor was present and no lymphadenopathy. Abdominal examination

revealed an intra-abdominal mass of about 10×12 cm in the left hypochondrium and epigastric region with extension into the umbilical region and the left lumbar region. The upper limit of the mass was not felt as it extended beyond costal margin. The mass was firm in consistency with a smooth surface. Other systemic examination was normal. The patient was admitted for further evaluation.

INVESTIGATIONS

Blood investigations revealed haemoglobin of 9.2 g/dL, erythrocyte sedimentation rate (ESR) of 80 mm in the first hour, the total leucocyte count and platelet count were within normal limits. Evaluation of anaemia using peripheral smear showed normocytic normochromic red blood cells and hypersegmented neutrophils; iron profile was suggestive of anaemia of chronic disease. Liver function and renal function tests, serum electrolytes, random blood glucose, serum amylase and lipase were normal. HIV antibody testing was carried out and the report was negative. Chest radiograph was also normal. Ultrasonography of the abdomen showed a large cyst of about 16×8×8 cm in relation to the tail of the pancreas and medial surface of the spleen possibly a pseudocyst of the pancreas. Contrast-enhanced CT (CECT) of the abdomen revealed a large well-defined lobulated cystic lesion of 16×10×8 cm arising from the upper and mid-pole of the spleen with no evidence of any solid component or calcification (figure 1). The lesion was found pushing the stomach posterolaterally towards right

Figure 1 Contrast-enhanced CT of the abdomen showing a well-defined lobulated cystic lesion arising from the upper and middle pole of the spleen with displacement of stomach posterolaterally.

CrossMark

To cite: Chakradhar K, Prasad S, Kumar S, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-203596

and abutting the anterior abdominal wall. Visualised portion of the head and the body of the pancreas was normal, and the tail of the pancreas was not well visualised. In view of CECT findings of splenic cyst, hydatid serology was performed, which was negative.

DIFFERENTIAL DIAGNOSIS

- ▶ Hydatid cyst of the spleen
- ▶ Pseudocyst of the spleen
- ▶ Pseudocyst of the pancreas
- ▶ Lymphoma of the spleen
- ▶ Metastasis to spleen

TREATMENT

The patient received vaccination against *Haemophilus influenzae* and pneumococci preoperatively. Laparotomy revealed a large cyst arising from the superomedial aspect of the body of the spleen. The cyst measured 16×10×8 cm almost involving majority of the spleen (figure 2). Superolaterally adhesions were present between the splenic cyst and the diaphragm. The rest of the abdominal viscera was normal. A total splenectomy was carried out as there was very little normal splenic tissue, which could be preserved. The patient had an uneventful, smooth postoperative recovery. Cytology of the fluid showed lymphocytes, an elevated adenosine deaminase (ADA) level of 79 IU/dL, an elevated lactate dehydrogenase of 3089 IU/dL and a low glucose level of 5 mg/dL. Histopathology revealed a fibrocollagenous wall devoid of living epithelium. The adjacent parenchyma showed extensive areas of necrosis and well-formed granulomas composed of giant cells, epithelioid cells surrounded by lymphocytes and plasma cells. The surrounding splenic parenchyma showed congested red pulp consistent with splenic TB with a pseudocyst (figure 3).

OUTCOME AND FOLLOW-UP

The patient was discharged on the fifth postoperative day. On 10th postoperative day, the patient was started on antitubercular treatment based on the histopathology report. The treatment regimen consisted of 6 months of treatment with four drugs (isoniazid, rifampicin, ethambutol and pyrazinamide) during the intensive phase and two drugs (isoniazid and rifampicin) during

Figure 2 Intraoperative photograph of splenic cyst arising from the upper pole of the spleen.

Figure 3 Image showing splenic tissue, granulomas and caseation necrosis suggestive of tuberculosis of the spleen on histopathological examination.

the continuation phase. Liver function tests were carried out 4 days after starting anti-TB regimen. The reports were within normal limits. The patient had come after 1 month for first follow-up. At this time it was seen that the patient had gained weight by 1 kg. The patient was again examined after 2 months and she was seen to be doing well. She had gained 2 kg weight and the ESR was 5 mm in the first hour. Liver function tests were within normal limits. Continuation phase of anti-TB regimen consisting of isoniazid and rifampicin was started.

DISCUSSION

Splenic TB is usually seen as a part of miliary TB most commonly in immunocompromised individuals.⁵ Isolated splenic TB is a very rare manifestation in immunocompetent individuals.² Although Winternitz⁶ categorised splenic TB as a primary or secondary form, most studies maintain that all patients with splenic TB are secondary to the previous infection of tubercle bacillus in other organs.⁷ Based on the pathomorphological features splenic TB can manifest in five forms including miliary TB, nodular TB, splenic abscess, calcific TB and the mixed type.⁸ Diagnosis of isolated splenic TB is usually delayed because of imprecise clinical manifestations. Splenic TB can be suspected on ultrasound abdomen as multiple small hypoechoic lesions and CECT abdomen may demonstrate hypodense lesions.⁸ Diagnosis is established by pathological examination of fine needle aspirate, splenic biopsy and splenectomy specimen.³ Suri *et al*⁹ reported up to 88% sensitivity for fine needle aspiration cytology for diagnosing tuberculous pathology in the spleen. Differential diagnosis of lymphoma, hydatid disease and metastasis should be considered. In the present case, the patient neither had a history of TB nor showed evidence of TB in any other organ, and hence can be regarded as isolated splenic TB.

Cysts of the spleen are rare lesions. Classification, diagnostic modalities and treatment guidelines are far from uniform. Three classification systems are present: Fowler,¹⁰ Martin¹¹ and Morgenstern.¹² Fowler¹⁰ is credited with the first classification system for splenic cysts. Martin¹¹ revised this to a more practical classification as type I and type II cysts. Type-I cysts are true cysts with cellular lining and may be of parasitic or non-parasitic origin. Non-parasitic type I cysts can be congenital or neoplastic. Type II cyst is a pseudocyst with no cellular lining. Morgenstern¹² proposed a classification of non-parasitic splenic cysts based on their macroscopic features into congenital,

neoplastic, post-traumatic and degenerative cyst. Pseudocysts that constitute 75% of all non-parasitic cysts are mostly of traumatic origin, but may also be of infectious and degenerative origin. Some of the causes are pancreatic pseudocyst in the spleen,¹³ secondary to mononucleosis from cytomegalovirus infection¹⁴ and subcapsular or intraparenchymal haematomas of the spleen due to acute or chronic pancreatitis.¹³ Pachter *et al*¹⁵ hypothesised that an increase in non-surgical management of blunt splenic trauma could contribute to the formation of splenic pseudocyst. Pseudocyst in tuberculous of the spleen is a rare entity.¹

In 30–45% of the cases splenic cysts are present as an asymptomatic painless abdominal mass.¹⁶ Splenic cyst usually manifests as vague abdominal pain, early satiety, nausea and vomiting. The non-specific symptoms, such as epigastric fullness or pain, may be due to compression of adjacent organs such as the stomach. Abdominal CECT is the diagnostic tool of choice as it can better delineate the trabeculated nature of the cyst wall and wall calcification. CECT of the abdomen has greater specificity in defining whether a lesion is cystic or solid.

Historically splenic cysts have been treated by splenectomy. With 0.2–4.3% risk of opportunistic post-splenectomy infections conservative procedures such as partial splenectomy with a stapler or harmonic scalpel,¹⁷ percutaneous aspiration and drainage, and laparoscopic fenestration of the splenic cyst have been tried as an alternative to splenectomy.¹⁸ Percutaneous drainage of splenic cyst results in high incidence of complications such as infection, cyst recurrence, bleeding¹⁹ and a dense inflammatory reaction around the spleen making subsequent surgical treatment much more difficult. Therefore, attempts at percutaneous aspiration or drainage of splenic cysts should be avoided. The possibility of recurrence and presence of an echinococcal cyst with a false-negative serology should be kept in mind while planning for splenic preservation surgery.¹ In the present case, due to the large size of the cyst which almost replaced the entire spleen splenectomy was carried out.

ADA is an enzyme of purine metabolism, which catalyses adenosine into inosine and is found in most human tissues particularly in the lymphoid tissues. This is an important enzyme in T-lymphocytes, where it is in a 10 times higher concentration than in erythrocytes. Its activity increases during the reproduction and response to antigenic stimulation of lymphocytes.²⁰ Therefore, its increased concentration may be found in all fluids occurring in the zones of TB serositis and hence used in diagnostics.^{21–22} The ADA level in tuberculous peritoneal effusion ranged from 35 to 135 U/L with a mean level of 92 U/L and a sensitivity and a specificity of 100% and 95%, respectively. In the non-tuberculous group, the ADA level ranged from 1 to 28 U/L.²¹ In the present case ADA levels in the pseudocyst fluid were 79 IU/dL consistent with TB.

Isolated splenic TB in a patient with no history of TB in the past should be considered as a new case of extrapulmonary TB according to the WHO guidelines.²³ The treatment regimen for these cases consists of 2 months of intensive phase therapy with four drugs—isoniazid, rifampicin, ethambutol, pyrazinamide and 4 months of continuation phase with two drugs, isoniazid and rifampicin.²³ Patient's body weight is used to monitor the response to treatment²³ in sputum negative pulmonary and extrapulmonary TB.

In the present case, the patient was started on anti-TB treatment with isoniazid (200 mg), rifampicin (400 mg), ethambutol (600 mg), pyrazinamide (1 g) for 2 months.²³ At the end of intensive phase, the patient had gained 2 kg weight and her ESR was 5 mm within the first hour. Then anti-TB regime was changed to isoniazid and rifampicin for the next 4 months.

Learning points

- ▶ Though uncommon, splenic tuberculosis can occur in immunocompetent hosts.
- ▶ Splenic tuberculosis can manifest itself as a pseudocyst of the spleen.
- ▶ Contrast-enhanced CT abdomen is the investigation of choice for the diagnosis of splenic pseudocysts.
- ▶ The diagnosis of splenic tuberculosis is established by culture of the abscess fluid, histopathological examination of the splenectomy specimen and by fine needle aspiration cytology.
- ▶ Anti-tuberculous regimen for isolated splenic tuberculosis should be given for 6 months (2-month intensive phase +4-month maintenance phase).

Contributors KC and SP are associated with the conception and design, acquisition of data or analysis and interpretation of data. KC, SK and MV are associated with drafting the article or revising it critically for important intellectual content. SP has contributed to the final approval of the version published.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Jain SK, Kaza RC, Vindal A. Pseudocyst of the spleen caused by tuberculosis: a rare entity. *BMJ Case Rep* 2009;2009:bcr06.2008.0102.
- 2 Sato T, Mori M, Inamatsu T, *et al*. Isolated splenic tuberculosis. *Nippon Ronnen Igakkai Zasshi* 1992;29:305–11.
- 3 Nayyar V, Ramakrishna B, Mathew G, *et al*. Response to anti tuberculous chemotherapy after splenectomy. *J Intern Med* 1993;233:81–3.
- 4 Dachman AH, Ros PR, Murari PJ, *et al*. Nonparasitic splenic cysts: a report of 52 cases with radiologic-pathologic correlation. *AJR Am J Roentgenol* 1986;147:537–42.
- 5 Prathmesh CS, Tamhankar AP, Rege SA, *et al*. Splenic tuberculosis and HIV-1 infection. *Lancet* 2002;369:353.
- 6 Winternitz MC. Tuberculosis of the spleen. *Arch Intern Med* 1912; 9:680–97.
- 7 Singh B, Ramdial PK, Royeppen E, *et al*. Isolated splenic tuberculosis. *Trop Doct* 2005;35:48–9.
- 8 Zhan F, Wang C-J, Lin J-Z, *et al*. Isolated splenic tuberculosis: a case report. *World J Gastrointest Pathophysiol* 2010;1:109–11.
- 9 Suri R, Gupta S, Gupta SK, *et al*. Ultrasound guided fine needle aspiration cytology in abdominal tuberculosis. *Br J Radiol* 1998;71:723–72.
- 10 Fowler RH. Cystic tumors of the spleen. *Int Abstract Surg* 1940; 70:213–23.
- 11 Martin JW. Congenital splenic cysts. *Am J Surg* 1958;96:302–08.
- 12 Morgenstern L. Nonparasitic splenic cysts: pathogenesis, classification and treatment. *J Am Coll Surg* 2002;194:306–14.
- 13 Agnifili A, Gianfelice F, Gola P, *et al*. A rare case of splenic hematoma following chronic pancreatitis—the diagnostic and therapeutic procedures. *Jpn J Surg* 1991;21:576–9.
- 14 Tosato F, Passaro U, Vasapolo LL, *et al*. Sub capsular splenic hematoma due to infectious mononucleosis. *Rev Eur Sci Med Farmacol* 1995;17:157–9.
- 15 Pachter HL, Hofstetter SR, Elkowits A, *et al*. Traumatic cyst of the spleen: the role of cystectomy and splenic preservation: experience with seven consecutive patients. *J Trauma* 1993;35:430–6.
- 16 Labruzzo C, Haritopoulos KN, Tayar AR, *et al*. Post-traumatic cyst of the spleen: a case report and review of literature. *Int Surg* 2002;82:152–6.
- 17 Uranus S, Kronberger L, Kraft-Kline J. Partial splenic resection using the TA-Stapler. *Am J Surg* 1994;168:49–53.
- 18 Sardi A, Ojeda HF, King D. Laparoscopic resection of a benign true cyst of the spleen with the harmonic scalpel producing high levels of CA 19–9 and carcinoembryonic antigen. *Am J Surg* 1998;64:1149–54.
- 19 Carpenter G, Cotter PW, Davidson JRM. Epidermoid cyst of the spleen. *Aust NZ J Surg* 1986;56:365–8.

- 20 Sharma SK, Suresh V, Mohan A, *et al.* A prospective study of sensitivity and specificity of adenosine deaminase estimation in the diagnosis of tuberculosis pleural effusion. *Indian J Chest Dis Allied Sci* 2001;43:149–55.
- 21 Gupta BK, Bharat V, Bandyopadhyay D. Sensitivity, specificity, negative and positive predictive values of adenosine deaminase in patients of tubercular and nontubercular serosal effusion in India. *J Clin Med Res* 2010;2:121–6.
- 22 Mathur PC, Tiwari KK, Sushima T, *et al.* Diagnostic value of adenosine deaminase (ADA) activity in tubercular serositis. *Indian J Tuberc* 2006; 53:92–5.
- 23 World Health organisation. *Treatment of tuberculosis guidelines [online]*. 4th edn. 2009:31–2. http://whqlibdoc.who.int/publications/2010/9789241547833_eng.pdf (accessed 28 Dec 2013).

Copyright 2014 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- Submit as many cases as you like
- Enjoy fast sympathetic peer review and rapid publication of accepted articles
- Access all the published articles
- Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow