Splenic pseudocyst with splenocutaneous fistula: Case report and review of literature.

Usman Mohammed Bello,[1] Callistus Nwachukwu,[1]NasiruAbdullahiIsmail,[1] Umar Mohammed Mukhtar[2]

[1]Department of Surgery Bayero University Kano/AminuKano Teaching Hospital, Kano; [2]Department of Surgery, Usman Danfodio University Teaching Hospital,Sokoto, Nigeria

Abstract

Splenic cyst with splenocutaneous fistula is extremely rare. It is mainly caused by trauma or parasitic infestation. Splenic cyst mainly present with right upper quadrant abdominal pain and swelling. It can be complicated by rupture however presentation as splenecutaneous fistula is extremely rare as only one case was reported in the literature.Management of splenic cyst can be partial or total splenectomy and can be done open or laparoscopic. In this case we present a case of splenic cyst with splenocutaneous fistula that was managed surgically with resection.

Introduction

Splenic cysts are cystic diseases of the spleen. They are generally rare and can be classified as parasitic and non parasitic.[1] Parasitic splenic cyst are mostly caused by *EchinococcusGranulosis*, while non parasitic splenic cyst can be primary (true, congenital), which are lined by epithelial covering (demoid, epidermoid ,mesothelia and endothelium, *i.e.*haemangioma), or secondary (pseudocyst, non epithelial), which are mainly post traumatic.[2] Although there are various reportsof splenic cyst in the literature only one casereport was presented with splenocutaneous fistula.[3] Other complications outlined in literature include haemorrhage, spontaneous rupture and susceptibility to trauma.[2] In this report we present a case of giantinfected splenic cyst with splenocutaneous fistula.

Case Report

A 60 years old woman presented with complaint of progressive left hypochondrial swelling of 7 years duration. A month prior to presentation the overlying skin said to have ruptured spontaneously discharging purulent and blood stained effluent. There was associated left hypochondrial pain, low grade non radiating and no known relieving or aggravating factors. There was also history of early satiety, weight loss but no history of vomiting, constipation or other gastrointestinal symptoms. There was history of occasional fever. She was earlier admitted in a peripheral hospital on account of symptoms of anaemia for which she had four pints of blood transfused. There was no history of trauma to the abdomen. Known hypertensive on drugs but not known diabetic.

General clinical examinations revealed amiddle aged woman, wasted, afebrile, with a tinge of jaundice. Abdominal examination revealedgrossly distended abdomen with obvious asymmetry. There was an ulcer at left hypochondrial region of about 8x6cm with a sinus opening in the middle of it discharging turbid effluent. A huge mass was palpated extending from the left hypochondrial region to left iliac fossa and umbilical region. Other examination findings were essentially normal. A clinical impression of pancreatic pseudocyst was made.

Abdominal CT scan showed a huge non enhancing oval shaped mass extending from left hypochondrial region to the pelvis (Figure 1), and a rim of calcification. There was displacement of the stomach, left kidney and great vessels to the right. Conclusion of splenic cyst with differential of pancreatic pseudocyst was made.

Upper gastrointestinal endoscopy shows a normal stomach however there was an extraluminal mass compressing on the stomach from the left side. Full blood count, urea, electrolytes and were essentially normal.

She had exploratory laparatomy with intra-operative findings of huge splenic cyst with fibrous adhesions to diaphragm, abdominal wall and pancrease. The stomach, colon and small bowel were all displaced to the right of midline. The cyst was collapsed with aspiration. About 3 litres of turbid fluid was aspirated. Total splenectomy was then done. There was inadvertent injury to pancreatic tail, and left hemi diaphragm due to anatomical distortion of the nearby structures and dense fibrous adhesion for which distal pancreatectomy and diaphragmatic repair with nylon suture was done. Chest tube was inserted and closed abdominal drain left in situ.The ulcerated skin in the left hypochondrial region was debrided serially and dressed and the ulcer healed subsequently (Figure 2). She did well and was discharged 3 weeks after the surgery. Histology of the resected spleen (Figure 3) shows features of splenic pseudocyst.

Correspondence: Usman Mohammed Bello,Department of Surgery, Bayero University Kano/Aminu Kano Teaching Hospital Kano, P.M.B 3411, Kano State, Nigeria
Tel.: +2348036893332
E-mail: belloum01@yahoo.co.uk

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Discussion

The earlier classification of splenic cyst into parasitic and non parasitic[1] has been modified by Martins.[4] He classified splenic cyst into Type 1 and Type II. Type I cyst are true cyst with epithelial lining. This can be parasitic and non parasitic. Non parasitic true cyst are congenital in origin, comprise 25% of splenic true cyst and commonly seen in children and young adults.[5] Although congenital splenic cyst has no malignant potential, however its associated with elevated CA 19-9 and CEA in many cases.[6-8] Splenic cyst of parasitic origin is caused by echinococcal species and mainly endemic in Africa, Latin America, middle

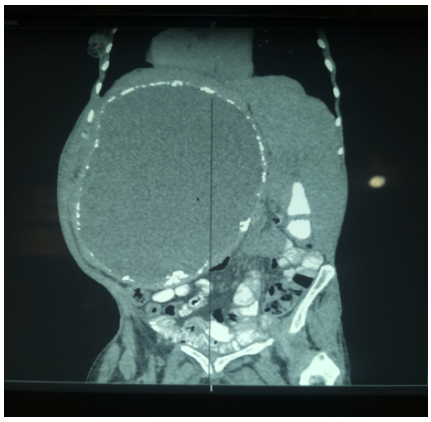

Figure 1. Abdominal CT scan showing the huge splenic cyst.

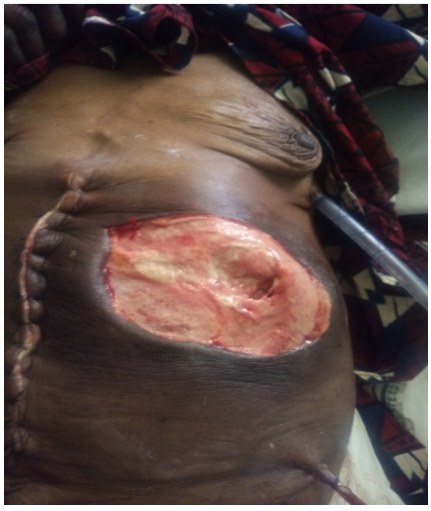

Figure 2. Debrided left hypochondrial skin showing the fistula opening after the splenectomy.

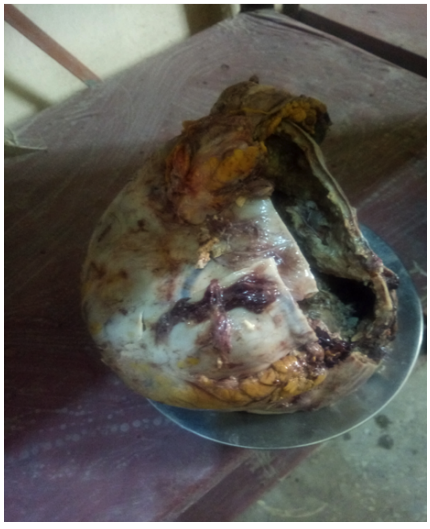

Figure 3. Resected splenic cyst after fixation in formaldehyde.

east and Australia, where there are domestic farming and cattle rearing activities. Spleen is the thirdmost common organ affected by hydatid disease. The most commonly involved organ is the liver (75%), followed by the lungs (15.4%), then spleen (5.1%).[9] In some instances simultaneous splenic and liver involvement have been reported.[10,11] Type II splenic cyst on the other hand is lined by granulation tissue and is mostly post traumatic in origin, following blunt abdominal injury.[12,13] The index case of pseudocyst however, history of trauma couldn't be recollected.

The clinical presentation includes pain in the right hypochonrial region, palpable mass in the right upper quadrantand markedly enlarged mass. There may be pressure symptoms of constipation or difficulty in breathing.[5-8] Rarely giant splenic cyst can erode the anterior abdominal wall and present as splenocutenous fistula as in this patient.[3] Investigation that confirm presence of splenic cyst are Abdominal CT scan, Abdominal ultrasound and occasionally endoluminal ultrasound scan and MRI.[4-6] In this patient abdominal CT scan done suggested splenic cyst with differential of pancreatic pseudocyst. The management of splenic cyst depends on the pathological type, size and location of the cyst.[14] Conservative approach is recommended for asymptomatic cyst that are less than 5cm in diameter as spontaneous resolution have been noted.[14] Various surgical options have been described and these include percutaneous drainage with or without injection of sclerosant, masupialisation, cyst fenestration, partialsplenectomy,and total splenectomy.[6,12,14] These procedures can be done open or laparoscopically. In our patient total splenectomy and distal pancreatectomy were done because the cyst has occupied the entire spleen and the contents were infected due to the fistula. Distal pancreatectomy was done due to distorted anatomy that resulted in inadvertent pancreatic tail injury. Complications that could arise during the surgery are usually due to extensive adhesions with surrounding structures in addition to the distorted anatomy. Organs to be injured during the surgery include stomach, colon, diaphragm, spleen and liver.[14] In our patient there was extensive adhesions which led topancreatic tail and diaphragmatic injury. Distal pancreatectomy was done with over running of the pancreatic stump with silk suture while the diaphragmatic injury was repaired with nylon suture.

Conclusions

In conclusion although splenic cyst with splenenocutaneous fistula is extremely rare, it should be suspected in a patient with left hypochondrial discharging sinus with concomitant left hypochondrial mass.

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