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A Rare Case Report of Splenic Cyst with Splenic Artery Aneurysm: An Incidental Combination

Dr. B. Sankararaman[(1)], Dr.NilakShantanurao[(2)] , Dr. V. Rajendran[(3)] , Dr.Kunduru Nava Kishore[(4)]

1. *Professor, Department of General Surgery, Vinayaka Mission Medical College and Hospital, Karaikal, India*
2. *2nd Year Postgraduate, Department of General Surgery,Vinayaka Mission Medical College and Hospital, Karaikal. India*
3. *Ex Professor and HOD, Department of General Surgery, Vinayaka Mission Medical College and Hospital, Karaikal, India*
4. *Associate Professor, Department of Surgical Gastroenterology, Nizam's Institute Of Medical Sciences, Hyderabad, Telangana, India*

Abstract:
Introduction:*Splenic cysts and splenic artery aneurysms (SAA) are rare, the combination itself is much rarer. Treatment is total splenectomy.*
Case report: *A 45-year female presented with abdominal distension since 2 years.She was diagnosed to be having large splenic cyst. Intra operativelya splenic artery aneurysm was found. Total splenectomy was done.*
Conclusion: *A large splenic cyst with splenic artery aneurysm has not been reported. Though various options of treatment of splenic cyst and SAAindividually reported, uniqueness of the combination in this case made total splenectomy the only treatment of choice.*
Key words: *splenic cyst, splenic artery aneurysm, total splenectomy***.**

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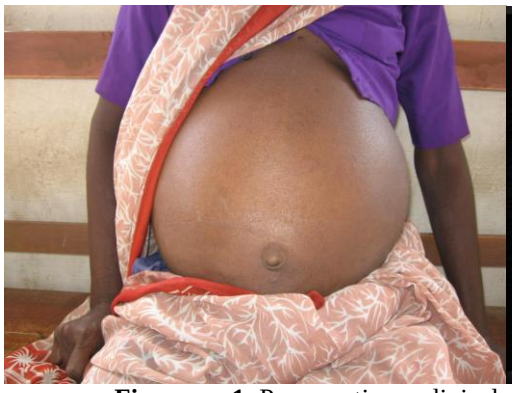
I. Introduction

Splenic cysts are rare; the reported incidence rate is 0.07% according to a large case series of 42,327 autopsies over a 25-year period (1), Splenic artery aneurysms occur in approximately 1% of the population and are usually incidental findings. Rupture of the aneurysm carries a substantial mortality (2). The incidence of SAA has been reported to be between 0.02% and 10.4% in the general population, with a predominant occurrence in multiparous women (3). Across the review of literature there has not been a case reported of co-presentation of a splenic cyst with splenic artery aneurysm. The aneurysm was an incidental intra operative finding, which was apparently looking like a cyst near splenic hilum on a plain CT of abdomen and USG.

II. Case report

A 45- year old female presented with complaints of abdominal distension and discomfort since two years. She had some difficulty in passing urine for 1 month, breathlessness for 1 month and abdominal pain for 10 days. Patient noticed a swelling in the left upper quadrant of abdomen two years back which increased in size gradually to attain the present size. History of anorexia, insomnia and low backache were present. Difficulty in passing urine was there (later on it was found to be due to cystocele). There was no H/O constipation or loose stools or melaena, fever or cough. There was no H/O abdominal trauma. She was not a known case of DM, TB, HTN. She had no pet dogs in her house. She was a non-vegetarian, non-alcoholic and non-smoker. She was treated at local hospitals for abdominal pain which was on and off. She was having regular menstrual periods. She was the mother of 3 children, all were born of full term normal deliveries. Last childbirth was 16 years back. On general examination the patient was poorly built and poorly nourished, was anaemic, afebrile. Vitals were within normal limits. No abnormality was found on examination of other systems. Per abdomen, it was soft and protuberant. Umbilicus was everted. No dilated veins were seen around umbilicus or flanks. A huge intra abdominal swelling was palpable extending from left hypochondrium through the left lumbar up-to the left iliac crest, also extending to right quadrants. Lower limit could not be defined (Figure no 1). Size was 25cm x 20cm, variable in consistency. Abdominal girth at the umbilicus was 110 cm. No free fluid was observable in the abdomen. Bilateral direct inguinal hernias were present. Bowel sounds were normal. Per rectal examination showed Rectocele. Per vaginal and per speculum examination showed third degree utero-vaginal prolapse, Cystocele, Enterocele, and Rectocele, Cervix was hypertrophied, did not bleed on touch. Blood investigations

revealed anaemia(6.6gm/dl). Renal functions and liver functions were within normal limits. Serology was negative. X-ray abdomen (Figure no 2) showed large soft tissue dense opacity in the center of abdomen, Splenic flexure and transverse colon were pushed upwards and ascending colon moved more to right. No abnormal calcification in Plain X Ray. CT-SCAN- Splenomegaly was noted. A large cyst was present on the lateral and anterior aspect of the spleen (Figure no 3). Septations were present in the cyst. Gall bladder stone (4mm) was noted incidentally. CEA < 1.0 µg/L (RR < 5.0), CA 19.9 was 120 ku/L (RR < 35), CA 125 was 11 ku/L (RR < 35), AFP 150 µg/L (RR 30–270), serum amylase and lipase were also within normal limits.



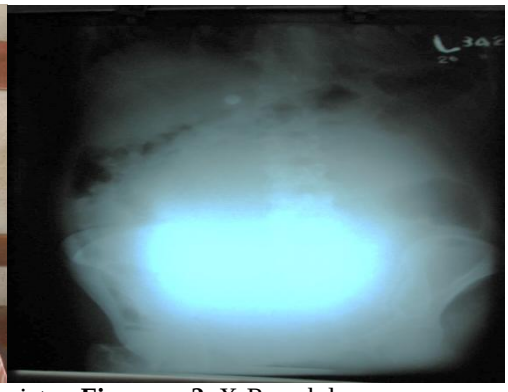


Figure no 1: Preoperative – clinical picture**Figure no2**: X-Ray abdomen

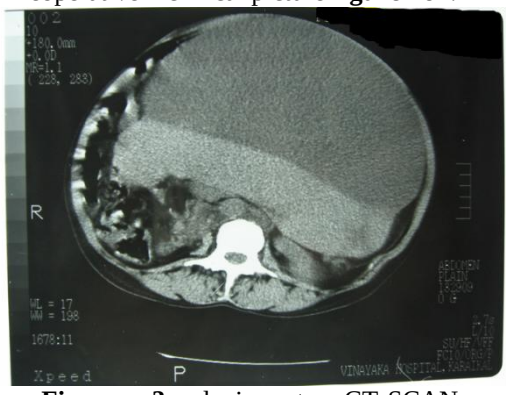


Figure no3: splenic cyst on CT-SCAN

TREATMENT-TREATMENT- Patient was taken up for emergency laparotomy as during the period of evaluation she developed sudden pain, with possible rupture. In supine position with General Anaesthesia, left paramedian Incision was made with T- cut onto the left side. Abdomen was opened and explored; intraoperative findings showed amber coloured fluid of 750 ml in the peritoneal cavity; Spleen was enlarged massively; Huge cyst on lateral and anterior aspect of spleen (ruptured at un-defined point) was found with similar fluid inside (Figure no 4). No adhesions were noted. Liver appeared cirrhotic. No evidence of portal hypertension was noted, such as dilated veins. Splenic artery aneurysm was present in the distal part close to the splenic hilum (Figure no 5). Ovaries and uterus were normal. Other viscera were normal. Spleno-renal ligament was opened, Short gastric vessels were secured with trans fixation sutures of silk. Splenic artery aneurysm clamped proximally and distally, and transfixed with 1-0 silk and divided distally. Splenic vein was transfixed and divided. Tail of pancreas was separated safely from the hilum of the spleen and the large spleen along with the cyst was delivered out of the abdomen (Figure no 6). A small wedge of liver was also taken for biopsy. Peritoneum was irrigated with saline. The incision was closed in layers. Intraoperatively 1 unit of whole blood was transfused. Post-operative care included antibiotics, analgesics and blood transfusions. Pneumococcal and H.influenza vaccines were given . Skin sutures were removed on the 12th day. Histopathological examination showed spleen with thickened capsule; red pulp seen with congested blood vessels; Gamma Gandy bodies were noted; cyst was containing mucinous material; there was no obvious lining in the cyst wall, suggesting Pseudocyst of spleen. The liver biopsy showed evidence of micronodular cirrhosis (? cause).

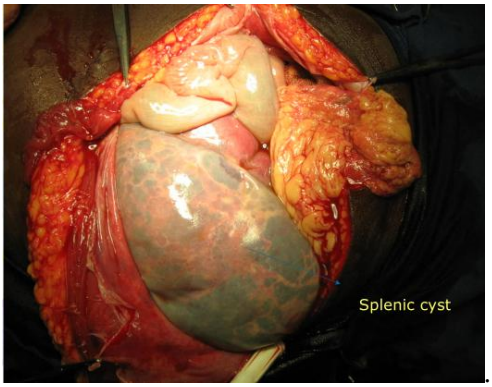


Figure no 4: Intraoperative picture 1

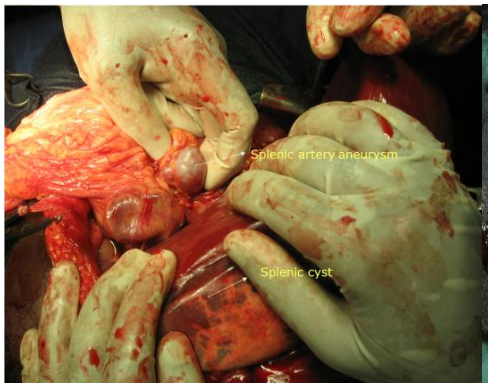


Figure no 5: Intraoperative picture 2

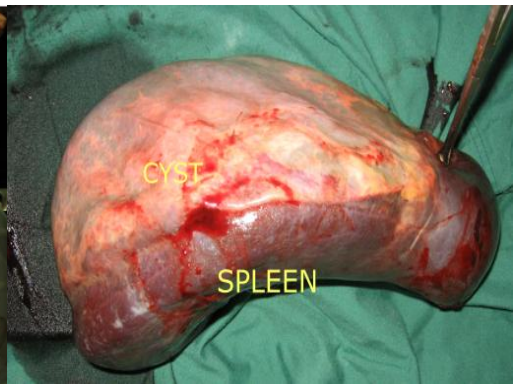


Figure no 6: Excision specimen

III. Discussion

Splenic cysts are most common in the second and third decades of life, although they have been noted in all age groups including infants. An asymptomatic abdominal mass is the presenting feature in 30% to 45% of cases. Abdominal symptoms such as pain may present in children and adults, but symptoms are more common when cysts are larger than 6 to 8 cm. The cyst may be accidentally noted during a physical examination of the patient. Symptoms and signs may arise from the compression on adjacent structures by the enlarging mass. Fowler described the first classification of splenic cyst. Splenic cysts are classified as primary or secondary cysts, according to the presence or absence of an epithelial lining. (Table no 1). The primary cysts are further subdivided as parasitic or non-parasitic. Secondary cysts are in most cases post traumatic.

Primary or true cyst
• Parasitic- echinococcal cyst
• Non-parasitic
○ Congenital- epidermoid, dermoid, endodermoid
○ Neoplastic- haemangioma, lymphangioma
Pseudocyst
• Post traumatic (Injury)
• Abscess (Infection)
• Haemorrhage (Infarction)

Table no 1: classification of splenic cyst[4,5,6,7]

Females are affected more [6], approximately two-thirds of all patients with splenic cysts are aged below 40 [10]. The majority of patients are either asymptomatic or experience minor, non-specific symptoms related to the mass effect of the cyst such as left upper abdominal pain, early satiety, nausea and vomiting [11]. Studies suggest that the inner cellular lining may be positive for CA 19-9 in immunohistochemistry. CA 19-9 serum level if elevated, returned to normal after resection of the cyst [15]. Imaging findings seen on computed tomography are septations, calcifications and internal debris with no enhancement [12]. Presence of multiple neighbouring cysts or daughter cysts, favours the diagnosis of parasitic cysts. Splenic cysts usually have a thin-walled anechoic appearance on ultrasound. There may be no evidence of calcification of the wall on computed tomography [13]. On T2-weighted magnetic resonance imaging, the cyst is hyper-intense, and on T1-weighted imaging, it is hypo-intense but can increase depending on cyst contents [14]. Treatment protocol depends upon

the final diagnosis. Echinococcal splenic cyst warrants total splenectomy. A congenital, benign cyst or a pseudocyst <5cm can be observed for a period of time; >5cm should be taken for splenectomy. Other options for treatment are percutaneous or laparoscopic aspiration or drainage with indwelling catheters. "Fenestration" involves resection of the wall of extra splenic cyst to create a permanent opening into the peritoneum, accomplished either by open or laparoscopic surgical techniques[16]. A better alternative involves partial splenic decapsulation, also known as marsupialization [16]. All modes of treatment other than total splenectomy need more studies to validate them, as they may not prevent recurrences. The incidence of OPSI (overwhelming post splenectomy infections) is reported to be 0.2% to 4.3%, with a lifetime risk of 5%. The most significant organisms responsible are Streptococcus pneumoniae, Neisseria meningitidis, and Haemophilus influenzae. Vaccines directed toward these three bacteria species are available in case a complete splenectomy is unavoidable [22].

An aneurysm is a localized abnormal dilation of an artery [17]. Any artery can be affected by a true or a 1pseudoaneurysm. In true aneurysm, the aneurysmal wall is composed of all the 3 layers, i.e., intima, media and adventitia. Whereas in a pseudoaneurysm there is a breach in the vessel wall along with an extravascular hematoma bounded by surrounding tissue. The most common site of intra abdominal aneurysm is the Abdominal Aorta followed by iliac arteries and then splenic artery [18]. Aneurysms in visceral arteries are rare but amongst them, splenic artery is the commonest site, being affected in 60% of cases [19]. The incidence of SAA in females is four times higher than in males. This difference is thought to be due to the hormonal and haemodynamic changes associated with pregnancy, though this is not seen with other visceral artery aneurysms [8]. Most patients with SAAs are asymptomatic [9]. An increased incidence of SAAs has been associated with several conditions, including pregnancy, degenerative atherosclerosis, portal hypertension, medial fibrodysplasia, arteritis, collagen vascular disease, α1-antitrypsin deficiency, and pancreatitis [20]. Pseudoaneurysms of the splenic artery are most often caused by chronic pancreatitis or by trauma [20]. SAAs are usually diagnosed incidentally. Diagnosis is usually made by contrast enhanced computed tomography scans wherein the lesions are imaged during the arterial phase. Doppler and magnetic resonance imaging are important adjunct in diagnosis. Digital subtraction angiography or transcatheter angiography via femoral artery is considered gold standard investigation. The risk of rupture in large aneurysms(>2cm) is as high as 28% [9] with mortality approaching 40% [21]. Traditionally, laparotomy with either aneurysm ligation alone or splenectomy with ligation was the treatment of choice of SAA. Today, various therapeutic options are available for SAA, including minimally invasive endovascular management and laparoscopic or open surgery with or without splenectomy [9].

Our case fits in the demographic pattern of both the pathologies namely, splenic cyst and SAA; the patient was a female of 45 years, with no history of trauma or pancreatitis. Though the management or the outcome of the condition did not change, this case deserves special attention because of the rare combination of the two pathological entities.

IV. Conclusion

Splenic cysts are rare benign lesions with pseudocyst of spleen being a further uncommon pathology. A splenic artery aneurysm coexisting with pseudocyst of spleen has not been reported so far in the literature. This is a rare combination with a high possibility of complications. Expeditious diagnosis and treatment are the cornerstones in such cases so as to prevent catastrophic events, such as rupture and subsequent mortality. Open surgery has been a standard treatment protocol and laparoscopic approach is an emerging option. For huge cysts of this size, open surgical approach is only possible.

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