'BURNT OUT' BENIGN SPLENIC CYST, MIMICKING INTRA-ABDOMINAL MALIGNANCY: CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT

The spleen performs important immunological function. Benign cysts, neoplasms and abscesses are identifiable cystic lesions of the spleen. Splenic cysts are very rare, and consist of Type 1 (parasitic) and Type 11 (non-parasitic) cysts. Very few cases of huge splenic cysts have been reported in literature. The most common symptoms are due to pressure effects on contiguous organs, causing pain, abdominal swelling and change in bowel habit. Management of these splenic cysts is controversial. Indications for surgical intervention, include symptomatic or large diameter cysts (>5cm).

We report a 57 year old lady with an 18 year history of recurrent left abdominal pain, progressive weight loss, easy satiety, and recurrent low grade fever. She neither had change in bowel habit, nor haematuria. There was no history of abdominal trauma. On physical examination, there was a left hypochondriac swelling, extending to the midline of the abdomen. She has been transfused severally in the past on account of recurrent anaemia. Ultrasound revealed multiple well circumscribed oval and rounded cysts of the spleen. Her haemoglobin level at presentation was 6g/dl. She had neutrophilia. She subsequently underwent total splenectomy with good surgical outcome.

KEYWORDS: *splenic cysts, recurrent anaemia, total splenectomy.*

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INTRODUCTION

Cysts of the spleen are rare lesions, with limited reports in the literature. It is rare in all age groups[1]. Splenic cysts can be parasitic or non-parasitic. *Echinococcus granulosus* infestation is the most common aetiologic organism for parasitic cysts of the spleen[2,3]. The non- parasitic cysts are further divided into primary (true, congenital, epidermoid/epithelial) or secondary (false) cysts depending on presence or absence of epithelial or endothelial lining. Primary splenic cysts are usually congenital and are incidentally diagnosed in early life. Secondary cysts are usually traumatic, lack epithelium, hence the name pseudocyst. Parasitic splenic

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cysts are more common in Africa and Central America, while non parasitic cysts are most common in Europe and North America[4]. These cysts are more common in children and adolescents, with female preponderance[3]. Splenic cysts are usually asymptomatic and often seen as incidental findings at imaging studies. Symptomatic splenic cysts result from enlargement, rupture, infection or haemorrhage[5]. The pathogenesis of primary cysts is believed to be either from entrapment of mesothelial cells of the peritoneum in the splenic parenchyma during embryogenesis or cysts originating from normal lymph spaces in the spleen[6]. Abdomino-pelvic ultrasonography and computed tomography scan are diagnostic. Management is usually controversial as the spleen is an immunological organ, and should be preserved when appropriate and feassible[7]. Preservation of at least 25% of splenic tissue gives protection against pneumococcal infection[5]. Treatment strategies aim at elimination of cysts and prevention of recurrence[8]. Splenic preservation is an acceptable treatment modality for blunt trauma to the spleen. However, treatment of choice for large symptomatic splenic cysts is splenectomy.

CASE REPORT

A 57 year old teacher presented to the clinic with an 18 year history of recurrent left upper abdominal pain /swelling, and a 2 week history of severe weakness. Pain was dull, non colicky, and never severe. Pain did not radiate to any other part of the body. There was no change in bowel habits. No melaena, haematochexia or haematuria. No history of abdominal trauma in the past. There was associated left abdominal fullness and progressive left upper abdominal swelling that was never generalised. She had easy satiety, progressive weight loss, with recurrent low grade fever. She has presented to many herbal homes and hospitals over the years, and has been transfused severally in the hospitals. Two weeks prior to presentation, she became severely weak and never improved despite repeated blood transfusions at a private hospital, hence the presentation for specialized care.

On examination, she was cachexic, severely pale and weak with a poor performance status. Abdominal examination revealed a mildly tender left hypochondrial mass that enlarged across the midline. No ascitis. Bowel sound was present and normoactive. Her haemoglobin at presentation was 6g/dl. WBC count was 10,700cells/mm^3 with neutrophilia (68%). Platelet count and clotting pofile were normal. Abdominopelvic

ultrasound revealed multiple well circumscribed oval and rounded cystic masses in the spleen. The spleen measured 25cm x 20cm x 9cm. She was resuscitated with IV fluids, six units of blood and antibiotics. A total splenectomy was done after optimisation. Intra-operatively, there were adhesion bands over the splenic mass and few pockets of abscess cavities. The splenic mass ruptured during mobilization and approximately 2000mls of non offensive straw colored effluent was suctioned. Post operative period was uneventful, and she was discharged on day 6 post operative day.

Resected splenic tissue (figure 1) was sent for histology. Macroscopy showed an organ that weighed 900g and measured 19.0cm x 15.0cm x 7.0cm. Cut section showed pockets of multi-locular cysts filled with seromucinous fluid, while in other area, there was a huge unilocular cavity with a rough floor and indurated base which was gritty to sectioning.

Microscopy showed splenic tissue with cavities lined by predominantly granulation tissue and scar tissue formations, with foci of cholesterol clefts, dystrophic calcification and numerous siderophages. No focus of abscess, specific inflammation or neoplasia was seen. Overall features were those of 'burnt out' benign cystic lesion of the spleen. Photomicrographs (figures 2 and 3) show dense chronic inflammation, numerous siderophages, lymphocytes, fibrosis and collagenization, with effaced architecture.

Patient has been seen thrice in follow up clinic visits. She is asymptomatic and has done so well. No more has easy satiety.

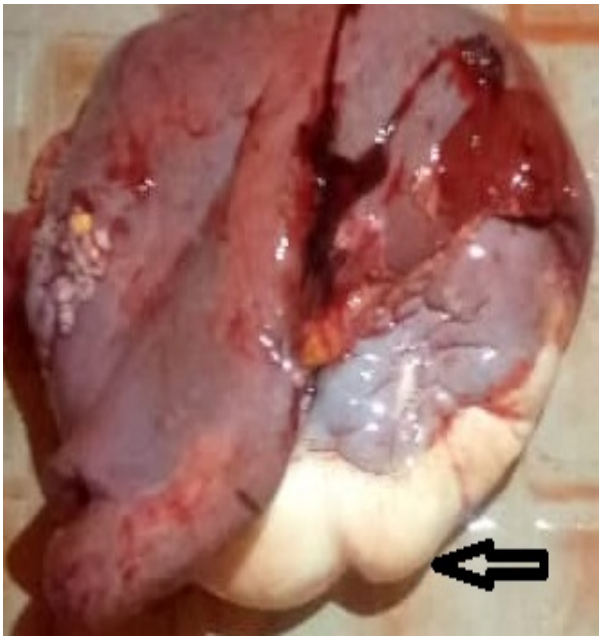

Figure1: Diaphragmatic (lateral) surface of resected huge splenic tissue, with arrow showing loculated cyst at the right inferior inferior border.

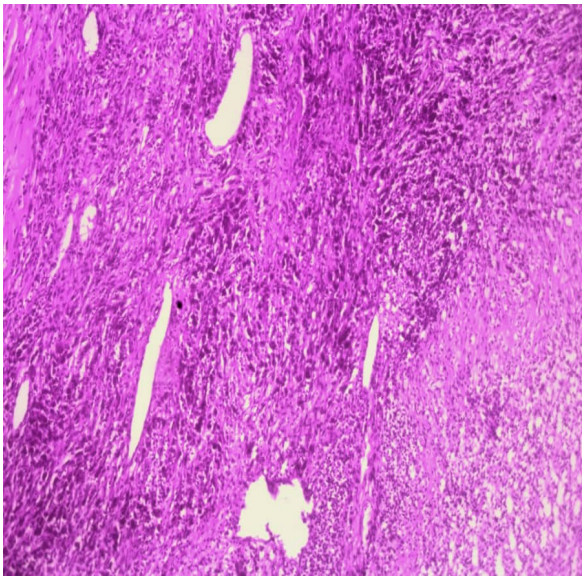

Figure 2: x100 photomicrograph showing siderophages, lymphocytes and fibrosis

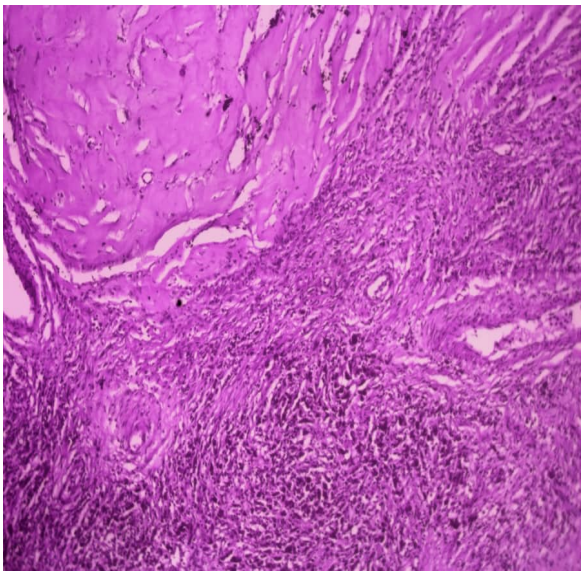

Figure 3: x100 photomicrograph showing chronic inflammation, with siderophages, fibrosis, collagenization and few vessels.

DISCUSSION

The spleen plays a significant role in hematopoiesis and immune-surveillance. It clears senescent and abnormal erythrocytes, opsonizes platelets and white cells as well as removal of microorganismsm and antigens. Also serves as a secondary lymphoid organ.

Huge splenic cysts result in asymmetric enlargement of the abdomen. The large cyst causes pressure effect on surrounding abdominal organs, notably the stomach and large intestine. The location and size of these cysts cause left hypochondriac pain, nausea, vomiting, anorexia and change in bowel habits[5]. Cysts larger than 5cm can rupture easily on minimal impact, and they have low probabilities of spontaneous resolution, hence need for prompt surgical intervention[9]. The differential diagnoses include parasitic disease, epidermoid/epithelial cyst, dermoid cyst, hemangioma, and/or lymphangioma[10]. Splinting of the diaphragm by large splenic cysts may result in breathlessness and discomfort to the patient[9]. Splenic cysts are more common in childhood and early adulthood with higher incidence in females[1,3,9]. Late diagnosis is possible in neglected or slow growing cysts as in the index case. The patient's age, sex, history of trauma and duration of symptoms are helpful in determining aetiology[11]. Our patient was 57 years old at presentation and has been symptomatic in the past 18 years. She has presented to many hospitals and herbal homes. She has taken a

lot of concoctions while at herbal homes. Notably, no scarification marks done. She had also been transfused severally at different hospitals, in the course of this illness. Recurrent anaemia results from intra-cystic bleeding, recurrent infections /inflammation (figures 2- 3) and /or poor appetite /easy satiety (leading to poor intake). Anemia can also result from benign lesions of lymphatic vessels and recurrent localized abscess formation following inflammatory reactions as seen intra-operatively in this patient. Boubacar et al[12] reported a case of isolated splenic lymphangioma in a 40 year old woman causing anemia and abdominal distension. Inflammatory pseudotumor causes fever, malaise and weight loss. Major complications of huge cysts are infection, rupture and/or bleeding. Spontaneous or traumatic rupture of large cysts is the most life threatening complication as a result of massive intra-abdominal haemorrhage[13].

Partial splenectomy is a recommended method for parenchymal preservation. However, total splenectomy is preferred for huge symptomatic splenic cysts to prevent recurrence and rupture with attendant haemorrhage[14]. Our patient underwent total splenectomy with excellent surgical outcome.

CONCLUSION

Splenic cysts are rare tumors that can mimick intra-abdominal malignancy. A detailed clinical evaluation and basic radiological imaging is of extreme importance in management of these lesions. Final diagnosis is histology. Total splenectomy for huge cysts prevents severe complications and guarantees good surgical outcome.

ETHICAL CONSIDERATION

Written informed consent was obtained from the patient, for the publication of this report and any accompanying images.

CONFLICTING INTERESTS

The authors declare that they have no conflicting interests

REFERENCES

1. B Farhangi, A Farhangi, A Firouzjahi, B Jahed. Huge epithelial nonparasitic splenic cyst: A case report and a review of treatment methods. Caspian Journal of Internal Medicine 2016;7(2):146-149
2. FC Brunicardi, DK Anderson, Billiar TR et al. Schwartz's principles of surgery. 8th Edition. Newyork: McGraw Hill;2005.1307-1308
3. A Rana, S Khichy, H Kaur et al. Large splenic cyst: A rare

presentation. Cureus 2021;13(4):e14435

4. GJ Sellers, PM Starker. Laparoscopic treatment of benign splenic cysts. Surgical Endoscopy 1997;11:766-768

5. G Meenu, J Deepika, K Brar, S Gupta, V Gupta et al. Large splenic cysts- A rare entity presenting as hydatid cyst. Human pathology: case reports 2017;10:96-97

6. FG Robbins, AE Yellin, RW Lingua, JR Craig, FL Turrill, WP Mikkelsen. Splenic-epidermoid cysts. Annals of Surgery 1978;187:231-235.

7. M Geraghty, IZ Khan, KC Conlon. Large primary splenic cyst: A laparoscopic technique. Journal of Minimal Access Surgery 2009;5(1):14-16

8. A Verma, A Yadav, S Sharma, D Saini, P Om et al.A rare splenic pseudocyst. Journal of Strength and Conditioning Research 2013;9:86-93

9. A Sadeghi, Z Naderpour, M Ebrahimpur, H Saffar. Non parasitic splenic cyst. Middle East Journal of Digestive Diseases 2017;9:242-243

10. P Mirilas, A Mentessidou, JE Skandalakis. Splenic cysts: are there so many types?. Journal of American College of Surgeons 2007;204;459-465

11. L Morgenstern. Nonparasitic splenic cysts: pathogenesis, classification and treatment. Journal of American College of Surgeons 2002;194:306-314

12. E Boubacar, G Atsame-Ebang, A Zabeirou, N Hammas.K Mazaz et al. Isolated splenic lymphangioma presenting as huge mass causing anemia and abdominal distension in an adult patient: a case report. Journal of Medical Case Reports 2018;12:97-99

13. G Fragandreas, S Papadopolous, I Gerogiannis, C Spyridis, D Tsantilas et al. Epithelial splenic cysts and life threatening splenic rupture. CHIRURGIA)2011;106:519-522

14. G Galyfos, Z Touloumis, K Palogos, K Stergios, M Chalasti et al. Oversized pseudocyst of the spleen: Report of two cases: Optimal management of oversized pseudocysts of the spleen. International Journal of Surgery Case Reports 2014;5(2):104-1.