

Mesothelial Splenic Cyst—A Case Report

V R Reddi,*^{MD}, M K Reddy,**^{MD}, B Srinivas,***^{MD}, C C Sekhar,*^{MD,DCH}, O Ramesh,****^{MS,MCh}

Abstract

A 26-year-old male presented with a left upper abdominal mass of one year's duration. Ultrasonography revealed a cystic lesion arising from the lower pole of the spleen. Total splenectomy was done and pathological examination of the cyst confirmed a true cyst with mesothelial lining without squamous metaplasia. The epithelial linings of these true cysts ranged from flattened low cuboidal, low columnar to squamous type and unilayered or stratified. The pathogenetic hypotheses as well as clinicopathological features of this rare lesion, which is usually found in children and young adults, were reviewed.

Ann Acad Med Singapore 1998; 27:880-2

Key words: Epithelial cyst of spleen, Non-parasitic splenic cyst, True cyst of spleen

Introduction

Non-parasitic cystic lesions of the spleen are unusual. They are classified essentially as primary (true, epithelial) and secondary (pseudo, non-epithelial) based on the presence or absence of lining epithelium. Among these, the primary ones are rare and appear as asymptomatic masses in the left hypochondrium.¹⁻³ The pathogenesis of true cyst is conjectural and numerous hypotheses are given by different authors.^{1,4-6} In 1929, Andral first described an epidermoid splenic cyst found at autopsy and Pean performed the first recorded splenectomy for cyst in 1867.⁷ We present a case of mesothelial (epithelial) splenic cyst with a brief literature review.

Case Report

A 26-year-old healthy male presented with a slowly enlarging mass in the left upper quadrant of the abdomen of one year duration with dull dragging pain. A history of injury to the abdomen due to a fall from a bicycle prior to the appearance of the mass was present. Physical examination revealed a non-tender tense mass with smooth surface in the left hypochondrium which moved with respiration. It was freely mobile from side to side and was 6 cm below the left costal margin. Spleen

could not be made out separately. Systemic examination, biochemical and haematological investigations were normal. Casoni's test was negative. Plain X-ray of the abdomen showed a large globular mass in the left hypochondrium and ultrasonography pointed to its cystic nature and origin from the lower pole of the spleen (Fig. 1) but no calcifications. Total splenectomy was accomplished and sent for histopathological examination. The postoperative period was uneventful and the patient was well 10 months after surgery.

The surgical specimen included a spleen with a large eccentrically located cyst weighing 800 g. The cyst measured 18 x 14 x 6 cm with intact surface, whereas the spleen was 6 x 3 cm size. On opening, the cyst was unilocular and contained about 500 ml of yellowish brown granular fluid. The inner surface was yellowish to whitish glistening with marked trabeculations and normal appearing adjacent splenic parenchyma (Fig. 2).

Microscopically, the fibrocollagenous cyst wall was lined by cuboidal to low columnar non-ciliated (mesothelial-like) epithelium, at areas stratified (Fig. 3). The epidermoid nature of the cyst was ruled out by the absence of squamous epithelium in numerous sections studied from different areas. The lining was ulcerated at

* Assistant Professor

** Additional Professor and Head

*** Senior Resident

Department of Pathology

SV Institute of Medical Sciences

**** Assistant Professor

Department of Surgery

SVRR Government Hospital

Address for Reprints: Dr V Ramana Reddi, Department of Pathology, Sri Venkateswara Institute of Medical Sciences, Tirupati-517 507, Andhra Pradesh, India.

E-mail: svims.nic.in/root

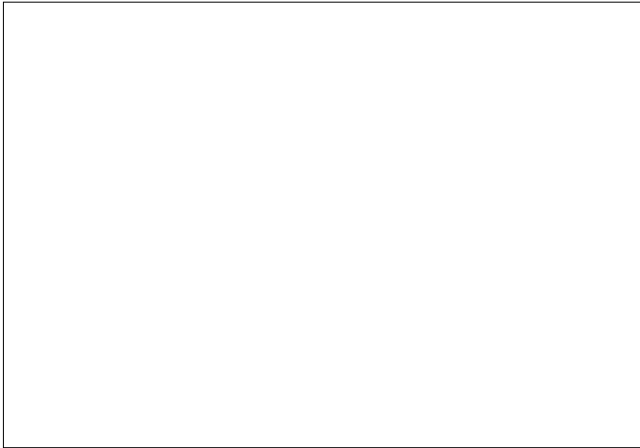


Fig. 1. Ultrasonography (abdomen) demonstrating a large cystic mass of splenic origin.

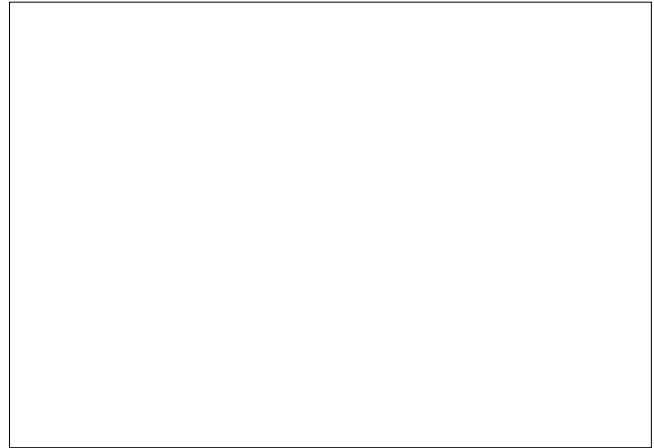


Fig. 2. Gross photograph depicting a large cyst with characteristic trabeculated inner surface and a part of normal spleen (arrow) to one side.

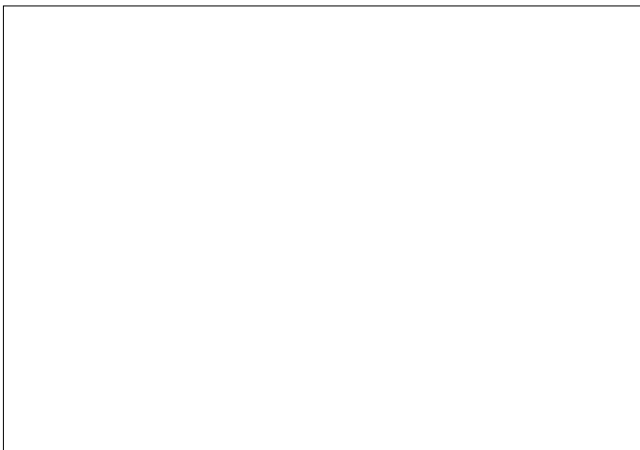


Fig. 3. Section of the cyst showing fibrocollagenous cyst wall with mostly unilayered cuboidal epithelium and a small stratified area (arrow). (H & E $\times 100$)

places covered by haemosiderin laden macrophages and mononuclear cells. Immunohistochemically, the epithelial cells were positive for cytokeratin (broad spectrum screening) and epithelial membrane antigen and negative for vimentin (DAKO, Denmark).

Discussion

Epithelial splenic cysts are rare which usually become symptomatic only when they attain a size of more than 5 cm in diameter.^{1,2} These cysts affect young people with a preponderance. Depending on the type of lining, these true splenic cysts are classified as angiomatous (endothelium), epidermoid (stratified non-keratinising squamous), simple congenital / mesothelial (low cuboidal to low columnar) and true dermoid (squamous lining with dermal structures) types.^{1,3-6} Among these, the endothelial lined cyst is thought to be different and considered as a cystic vascular lesion which is composed of several ectatic vessels, and would be more properly referred to as lymphangioma or haemangioma.⁵ Exclud-

ing these vascular cysts, the remaining constitute the true epithelial cyst where the epidermoid cyst is the most common one. Some prefer to call the epidermoid cysts as "mesothelial cysts with focal squamous metaplasia", because in most cases the squamous epithelium is focal and continuous with the mesothelial lining.⁵

There is still controversy regarding aetiopathogenesis of these uncommon lesions. The primary cysts are thought to be congenital and have been attributed to developmental misplacement of epithelial tissue during embryogenesis with consequent metaplasia.¹⁻⁴ But Ough and others⁵ suggested that these cysts excluding dermoid cysts originate from the invagination of splenic capsular mesothelium during development with consequent fluid accumulation for unknown reasons, resulting in cyst formation. By doing immunohistochemical and morphological investigations, including electron microscopy, Burrig⁶ concluded that most epithelial cysts are derived from traumatic damage to the splenic capsule with resultant ingrowth of surface mesothelium and subsequent cyst formation. As in our case, a history of previous abdominal trauma has been noted in most of the reported cases and found that it was the most important factor in cyst development. But some, doubted its aetiopathogenetic association, and concluded that injury leads to haemorrhage in an already existing cyst, resulting in clinical enlargement.^{1,2,8} The occurrence of various types of epithelium in true cysts may simply be explained by implicating the pleuripotent nature of mesothelial cells.^{5,6} By doing immunohistochemistry with a battery of monoclonal antibodies to cytokeratins and comparing it with other squamous epithelia, Lifschitz - Mercer et al⁶ suggested that the epidermoid cyst is either of teratomatous derivation or originates from inclusion of foetal squamous epithelium rather than from squamous metaplasia of mesothelium.

The cystic nature and splenic origin of these lesions

can be well established by various radiological studies such as ultrasonography, computed tomographic scan and magnetic resonance imaging.^{2,3,7} Though radioisotope splenic scan and selective angiography are reported as being the most certain means of diagnosing the cyst as benign, they are usually unnecessary because the final characterisation depends on the lining of the cyst. "It is necessary to look carefully for the lining of the cyst, in case this is not found in the first section."^{1,4} Though the cysts are asymptomatic, except for the mass and pain in the abdomen, they can become infected or ruptured causing acute abdominal emergency.^{2,3,5,6} The cysts are usually unilocular with characteristic gross features and sometimes calcified. Though no associated cysts are noted in other viscera, a few case reports on familial true splenic cysts were recorded.⁹ Total splenectomy is usually performed for splenic cysts, but spleen saving surgeries such as partial splenectomy, marsupialization of the cysts and TA-Stapler usage¹⁰ are preferred because of the risk of severe post splenectomy sepsis.

REFERENCES

1. Tsakraklides V, Hadley T. Epidermoid cysts of the spleen. *Arch Pathol* 1973; 96:251-4.
2. Qureshi M A, Hafner C D. Clinical manifestations of splenic cysts: study of 75 cases. *Am Surg* 1965; 31:605-8.
3. Cave R H, Garvin D F, Doohen D J. Metaplastic mesodermal cyst of the spleen. *Am Surg* 1971; 37:97-102.
4. Rappaport H. Tumours of haemopoietic system. *Atlas of Tumour Pathology. Fascicle 8*. Washington DC, Armed Forces Institute of Pathology, 1966:388-96.
5. Ough Y D, Nash H R, Wood D A. Mesothelial cysts of the spleen with squamous metaplasia. *Am J Clin Pathol* 1981; 76:666-9.
6. Burring K F. Epithelial (true) splenic cysts. *Am J Surg Pathol* 1988; 12:275-81.
7. Schwartz S I. The spleen. In: Schwartz S I, Harold E, editors. *Maingot's Abdominal Operations*. Vol II. 9th ed. Connecticut, USA: Appleton & Lange, 1990:1679-80.
8. Lifschitz-Mercer D, Open M, Kushnir I, Czernobilsky B. Epidermoid cyst of the spleen: a cytokeratin profile with comparison to other squamous epithelia. *Virchows Arch* 1994; 424:213-6.
9. Welton C A, Sijbrandij E S. Familial non-parasitic splenic cysts [abstract]. *Neth J Med* 1992; 40:236-9.
10. Uran U S, Kronberger L, Neumayer K, Beham A. TA-stapler resection of congenital splenic cyst - case report [abstract]. *Acta Chir Scand* 1990; 156:247-9.