CASE REPORT

Primary Splenic Cysts: Report of Two Cases

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ABSTRACT

Non-parasitic primary (epidermoid) splenic cysts are rare lesions and manifest clinically only when they attain large sizes. Though benign in nature, yet early removal of splenic cysts is essential to avoid serious complications. Two such cases of primary splenic cysts are being presented. Splenectomy was required in both the cases because of their large sizes. On histopathological examination both were found to be primary (epidermoid) splenic cysts but each with a distinctive feature. In the first case, in addition to the large cyst and multiple microscopic cysts were also seen in the adjacent areas. While in the second case, the importance of a diligent search for the lining epithelium is highlighted as the stratified squamous epithelial lining was identifiable only in few foci while in most areas; there was ulceration and organization of exudates.

KEY WORDS: Cyst, epidermoid, primary, spleen.

Introduction

Non-parasitic cystic lesions of the spleen are unusual, of which the primary/epithelial cysts are rare.[1,2] Various hypotheses have been postulated regarding its etiopathogenesis, and over the years its classification too continues to evolve.[3,4] The cysts manifest clinically only when they attain large sizes. Although ultrasound and computed tomography (CT) scan can establish their cystic nature, only after microscopic examination can one subtype it as a true or false cyst.[1-3] Two cases of primary splenic cysts are being presented.

Case Report

A 16 years boy (Case 1) and 21 years female (Case 2) presented with similar complaints of dull aching pain and palpable lump in the left upper quadrant of abdomen of 6 months and 10 months duration, respectively. In both cases, ultrasonography and CT scan detected enlarged spleen with the large well-defined cystic lesion and were reported as splenic cysts. Splenectomy specimens of both cases were sent for histopathological examination and measured 16 cm × 11 cm × 9 cm and 12 cm × 9 cm × 8 cm, respectively. When cut open, both the spleens revealed unilocular cysts measuring 13 cm × 9 cm × 5 cm and 9 cm × 6 cm × 5 cm, respectively. Compressed splenic tissue could be identified at the periphery of the cysts. The cysts contained dark fluid, and the inner surface was pale glistening with marked trabeculations (Figure 1).

Microscopically, in case 1 the cyst was lined by flattened to cuboidal epithelial cells that were continuous with large areas that showed stratified squamous epithelium. Underlying the epithelial lining was a thick layer of dense fibrocollagenous tissue. Smaller cysts with similar epithelial lining were seen within it and adjacent splenic tissue (Figure 2). Immediately beneath, the wall was blood vessels with a thick rim of fibrous tissue (Figure 3). Foci of hemorrhage, granular necrotic debris, and cholesterol clefts were also seen (Figure 1). Sections from adjacent splenic tissue showed congestion.

In case 2, microscopic examination of initial sections taken from different areas of the cyst revealed only a fibrocollagenous wall with no lining epithelium. More sections were studied, and a stratified squamous epithelial lining could be identified only in few foci (Figure 4). Remaining features were very similar to case 1, except that no additional cysts were found in this case.

Both cases were reported on histopathological examination as Non-parasitic primary (epidermoid) splenic cysts.

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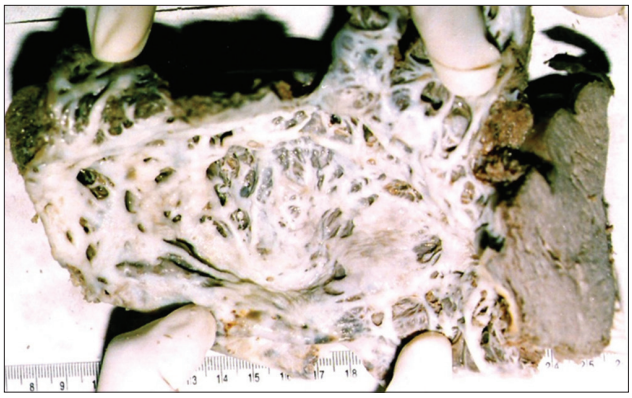

Figure 1: Cut open cyst showing pale glistening inner surface with marked trabeculations

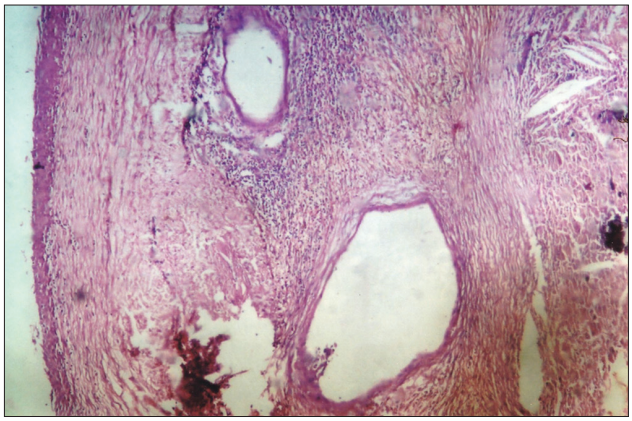

Figure 2: Stratified squamous epithelial lining with underlying dense fibrocollagenous tissue and scattered smaller cysts with similar epithelial lining. Granular necrotic debris and cholesterol clefts also seen (H and E, ×200)

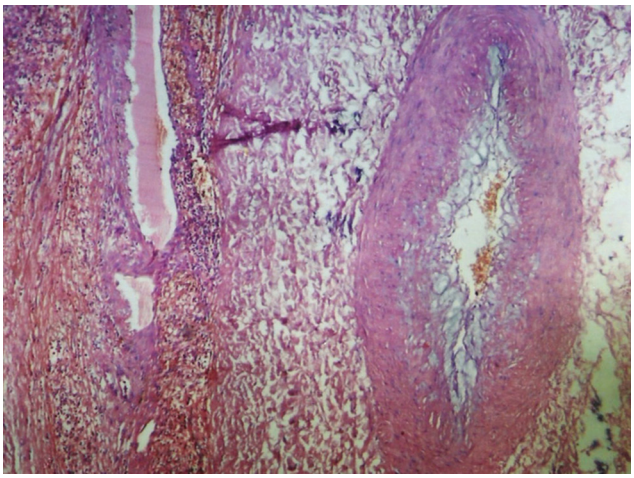

Figure 3: Blood vessels lying immediately beneath the wall with surrounding fibrous tissue (H and E, ×200)

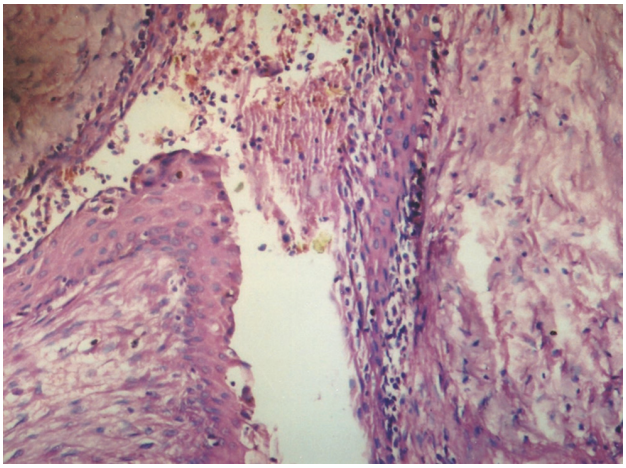

Figure 4: Stratified squamous epithelial lining could be identified only in few foci (H and E, ×400)

Discussion

Splenic cysts can be classified as parasitic (due to Echinococcus granulosus) and non-parasitic, of which the non-parasitic cysts are quite uncommon. The non-parasitic cysts can be primary (true/epithelial) and secondary (false/pseudo) depending on the presence or absence of an epithelial lining. Of these, true cysts are rare while secondary cysts form the major bulk (75%) of cases. Based on the cyst wall lining, the primary cysts can be subtyped into epidermoid (stratified squamous lining without adnexal structures), dermoid (squamous lining with adnexal structures), mesothelial (cuboidal to low columnar), and angiomatous (endothelium).[2-5] Of these, most authors prefer to refer to the endothelial-lined cysts as lymphangioma or hemangioma rather than include them as true splenic cysts.[4,5] Of the remaining primary cysts, epidermoid cysts are comparatively more common than mesothelial cysts.[4,5] Fowler introduced the term epidermoid cyst, but others prefer to refer to cysts lined by stratified squamous epithelium as Mesothelial cysts with squamous metaplasia or metaplastic mesodermal cysts.[4,6] However, the classification of splenic cysts continues to evolve with few authors expressing the view that lining alone cannot be a reliable criterion for classifying the cysts.[1,7] The pseudocysts are usually secondary to trauma, hemorrhage, infection or infarction and have a fibrocollagenous wall.[2,3]

There are various hypotheses regarding the etiopathogenesis of primary splenic cysts. Some consider it as developmental misplacement of

epithelial tissue during embryogenesis with consequent metaplasia while others suggest that the epidermoid cysts are congenital and either of teratomatous origin or derived from inclusion of fetal squamous epithelium rather than metaplasia.[4-6] Yet others are of the opinion that the invagination of capsular surface mesothelium during development and fluid accumulation within it may be the cause of cyst formation. While the pluripotent nature of the mesothelial cells explains, the variety of the lining epithelium that can be seen. The mesothelial nature of the lining cells is supported by immunohistochemical and electron microscopic findings.[3,4]

Abdominal trauma that has been noted in many cases has been given importance as a causative factor in splenic cyst formation and development.[3,5,7] However in both, the present cases no history of any abdominal trauma was obtained. This favors the suggestion made by some authors that abdominal trauma leads to intra-cystic hemorrhage from ectatic vascular channels, and contributes to the growth of the already existing cyst.[6,8] Blood vessels were also seen in the present cases as shown in Figure 3. The hemorrhagic nature of the cyst contents has been attributed to stoma-like sub-epithelial vascular channels that provide connection between the lumen of the cyst, and adjacent splenic sinuses and are vulnerable to trauma and pressure.[4,6] Primary splenic cysts are more common encountered in children and young adults and become symptomatic only when they attain large sizes.[4-6] The patient with splenic cyst can present with local (abdominal) or referred (left shoulder) pain, symptoms related to splenomegaly, atelectasis, occasionally infection, abscess formation and even rupture, and hemorrhage.[2,9]

Primary splenic cysts are usually unilocular, and occasionally satellite or daughter cysts may be seen lying adjacent to the main cyst and even communicating with it. This could be a result of invaginations of the surface epithelium of the main cyst.[4] Such satellite cysts were seen in the case 1 lying just below the epithelial lining as shown in Figure 2, but communication could not be identified. The trabeculations on the inner surface of the cyst (Figure 1) is thought to be due to the organization of stromal hemorrhage and resultant subepithelial fibrosis.[4,6] Though a diagnosis of splenic cyst can be established by radiological means, yet histopathology is mandatory to determine whether it is primary or secondary. Moreover, a diligent search should always be made for the cyst wall lining as its desquamation is known, which may lead to an incorrect interpretation of secondary cyst.[5-7] Elevated CA 19-9 serum levels accompanying primary splenic cysts have also been reported.[9]

Though benign in nature, yet surgery is essential to avoid serious complications such as rupture or infection leading to an acute abdominal emergency. Previously, splenectomy was the accepted treatment for splenic cysts. However with the realization of the importance of spleen in immunologic defense, currently more conservative surgery is preferred wherever more than 25% of splenic parenchyma preservation is possible.[2,3] However, due to increased risk of complications and recurrence, surgery is indicated in cases of very large or multiple cysts, cysts involving splenic hilum, and cysts adherent to adjacent structures.[2,8]

To conclude, primary splenic cysts are rare lesions, and this was re-emphasized when a search of literature provided only single case reports and small case series. Splenectomy was required in both the cases presented here because of their large sizes. In the first case, in addition to the large cyst, multiple microscopic cysts were also seen in the adjacent areas, which could have probably led to recurrence if splenic parenchyma saving surgery had been performed. The second case highlights, the importance of a diligent search for the lining epithelium as desquamation of the lining can lead to a misinterpretation of the pseudocyst.

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