

Letter to the Editor

Post-traumatic splenic cyst presenting as epidermoid cyst in spleen: an unusual presentation

Sir,

Epidermoid splenic cysts are very rare among primary splenic cysts. The histologic findings show cyst wall lined by stratified squamous epithelium, various theories have emerged regarding the origin of cysts, however controversies still exist. We report a case of young adult male in his 20s presenting with left upper abdominal pain and distention treated with open surgical procedure. Histopathology and immunohistochemistry findings further confirming the diagnosis as epidermoid splenic cyst. The case is unique in its association with history of trauma and development of true cyst.

A young adult patient in his 20s presented with history of left upper quadrant abdominal pain since 10 days visited a gastrosurgery OPD. The patient had history of blunt trauma to the abdomen 2 years ago with history of abdominal pain and swelling in left upper quadrant of abdomen since then. On examination, abdominal palpation demonstrated a non-tender and non-mobile lump in the left upper quadrant. Computed tomography (CT) scan showed a large, lobulated, thin walled cystic lesion in the spleen measuring approximately 20×20×18 cm in size without any evidence of a intralesional soft tissue component (Figure 1a). The patient was planned for laparoscopic surgery which was converted to open splenectomy mid procedure. The resected specimen was subsequently sent for histopathological examination. Specimen of splenectomy measuring 12×10×7.2 cm in size was received. Outer surface appears congested and shiny. Cut surface showed a unilocular cyst, showing trabeculations and yellowish to brownish exudate on inner surface (Figure 1b and c). Sections showed cyst lined by partly denuded stratified squamous epithelium with focal areas showing cuboidal lining. The cyst wall showed marked fibrosis, hyalinization and areas of calcification (Figure 1d-f). Splenic parenchyma adjacent to the cyst wall showed sinusoidal congestion and splenic white pulp comprising of residual lymphoid aggregates. Immunohistochemistry for Pan cytokeratin, cytokeratin 5/6 and p63 highlighted the squamous epithelium (Figure 1g-i) and diagnosis of epidermoid cyst of the spleen was rendered.

Splenic cyst presenting as large cystic mass is rare, the most common etiology is trauma which usually presents as pseudocyst. In the present case the splenic mass was preceded by history of trauma which might be the cause of the development, however the cyst wall was lined by

stratified squamous epithelium as true cyst while post traumatic cyst had no lining epithelium, which leads to the developmental dilemma. In the literature it is stated that the true cyst (epidermoid cyst) develops during the embryogenesis due to invagination of the parietal peritoneum supporting metaplasia theory from mesothelium to squamous epithelium. In the studies by Touloukian et al and Lee et al proposed and supported the possibility of development of splenic epithelial cysts from injury which leads to fluid collection or after spontaneous splenic bleeding, supporting the phenomenon of immature metaplasia which is supported by immunohistochemistry finding showing positivity for keratins, CEA while showing negative expression for BerEp4 as in the present case.^{1,2}

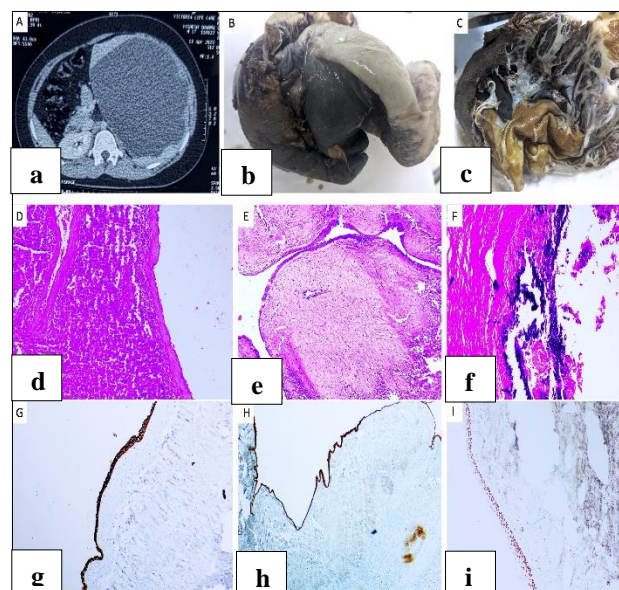


Figure 1: Radiologic and gross findings: (a) CT scan finding revealed an enlarged spleen with large thin walled cystic lesion measuring 20×18×20 cm in size showing minimal wall enhancement, (b) gross image shows cystic mass with compressed splenic parenchyma, (c) cut surface showing marked pearly white trabeculation and brownish yellow deposits on the surface, (d and e) histopathologic findings - photomicrograph showing cyst wall lined by partly denuded and partly by stratified squamous epithelium, wall showing, (f) areas of calcification, and (g-i) showing Pan CK, CK5/6 and p63 highlighting the lining epithelium (Hematoxylin and eosin original magnification (a-c) 100x, (d-f) DAB 200x).

However, study by Lifschitz et al proposed that the epidermoid splenic cyst is either of teratomatous derivation or originates from inclusion of fetal squamous epithelium after observing the cytokeratin (CK) profile and comparing the CK profile of epidermoid splenic cyst with stratified squamous epithelium of ovarian mature cystic teratoma and of fetal epidermis.³

In the present case the lining epithelium showed positivity for pan keratin, CK5/6 and p63 while negative for calretinin and Ber-Ep4 which is in accordance with the study by Touloukian et al supporting the fact that epidermoid and mesothelial cyst had distinct origin as opposed to Burring's theory which supports the fact the epidermoid splenic cyst seems to be a variant of the mesothelial cyst with focal squamous metaplasia.⁴ Various theories exist regarding the development of the epidermoid splenic cyst, however controversies still exists.

Surgery is the gold standard, including partial or complete splenectomy or partial cystectomy or decapsulation, the choice of surgery largely depending upon the age of the patient and the size, position and nature of the cyst. With the advent of laparoscopic surgery, it is the preferred method, since it leads to better recovery from pain, with less duration of hospitalization as compared to open procedure, moreover it offers better cosmesis.⁵

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