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CASE REPORT

A rare case of mucinous cystadenoma of the spleen in Libya

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ABSTRACT

This is a case of benign mucinous cystadenoma of the spleen in Benghazi, Libya. It is the first reported in Libya and most probably the fourth in Africa. Primary mucinous cystadenoma of the spleen is an extremely unique benign cystic lesion. Only a very few number of cases have been reported. These cysts are assumed to arise from invagination of the splenic capsular epithelium or from the ectopic pancreatic tissue. We report a rare case of primary mucinous cystadenoma of the spleen without evidence of pancreatic tissue in the specimen. Despite being a rare condition, mucinous cystadenoma of the spleen should remain in the differential diagnosis of any splenic cysts.

Keywords: splenic cysts, cystadenoma, Mucinous

INTRODUCTION

Primary mucinous cystadenoma of the spleen is an extremely unique benign cystic lesion that commonly affects the ovary, pancreas, and appendix.[1] Other unusual sites, such as the spleen, lung, urinary bladder, liver, terminal ileum, and retroperitonium, have been reported.[2] This type of benign tumor has rarely been reported in the spleen. The exact mechanism of splenic mucinous cystadenoma is still unknown, except for few situations, which either arise from an ectopic pancreatic tissue within the spleen or is associated with pseudomyxoma peritonei or mucocele of the appendix. More also, invagination of the splenic capsular epithelium is also assumed as another cause of this rare benign tumor.[3–7] However, splenic tumors are classified into four different categories: lymphoid, non-lymphoid and tumor-like lesions, and metastatic tumor. The tumor-like lesions include cysts and hamartomas.[4] In this report, we introduce a rare case of primary mucinous cystadenoma of the spleen in an elderly Libyan woman. Informed consent was obtained

from the patient regarding the images and other clinical information to be reported in this article.

CASE REPORT

A 74-year Libyan housewife presented symptoms of left upper abdominal pain for 4 months. Physical examination showed left hypochondrial tenderness with no palpable organs. Computed tomography (CT) scan of the abdomen revealed multiple variable sizes of cystic lesions arising from the spleen, which displaces the left kidney posteriorly. These lesions had a low attenuation with faint enhancement. Some of

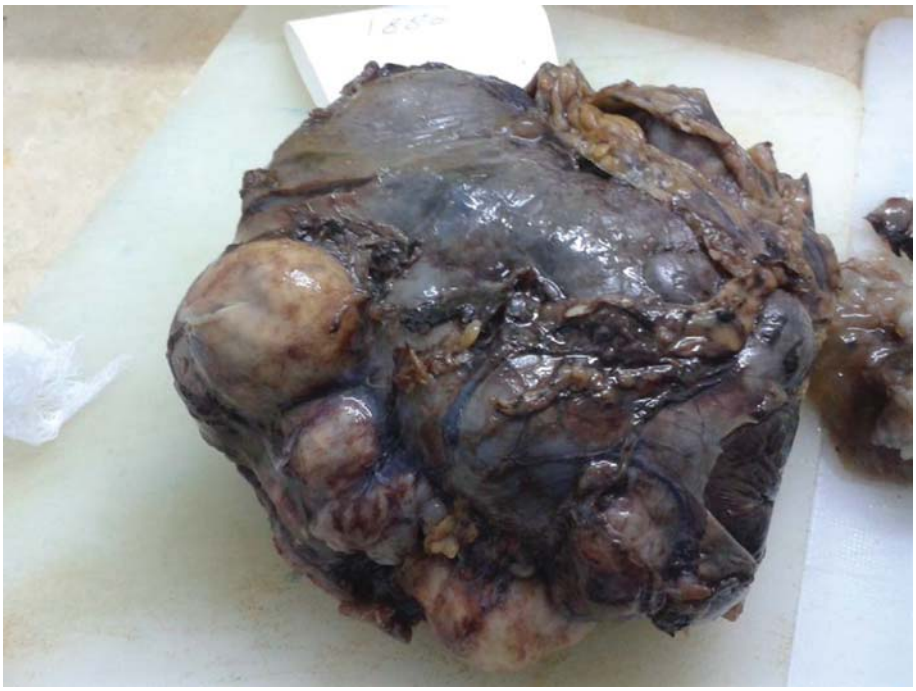


Figure 1. Appearance of the spleen after removal showing the multiple variable sizes cysts.

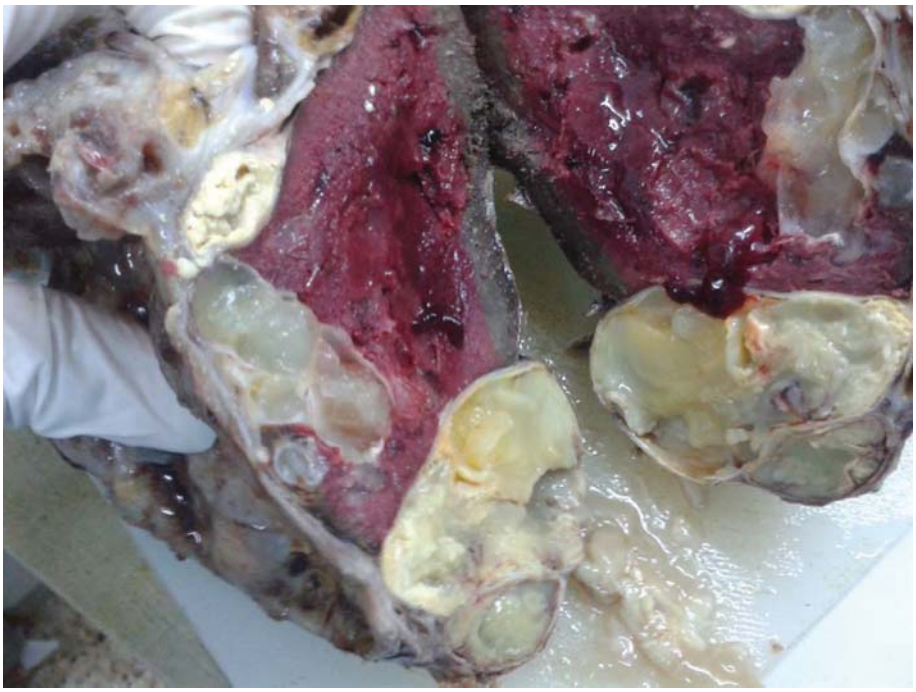
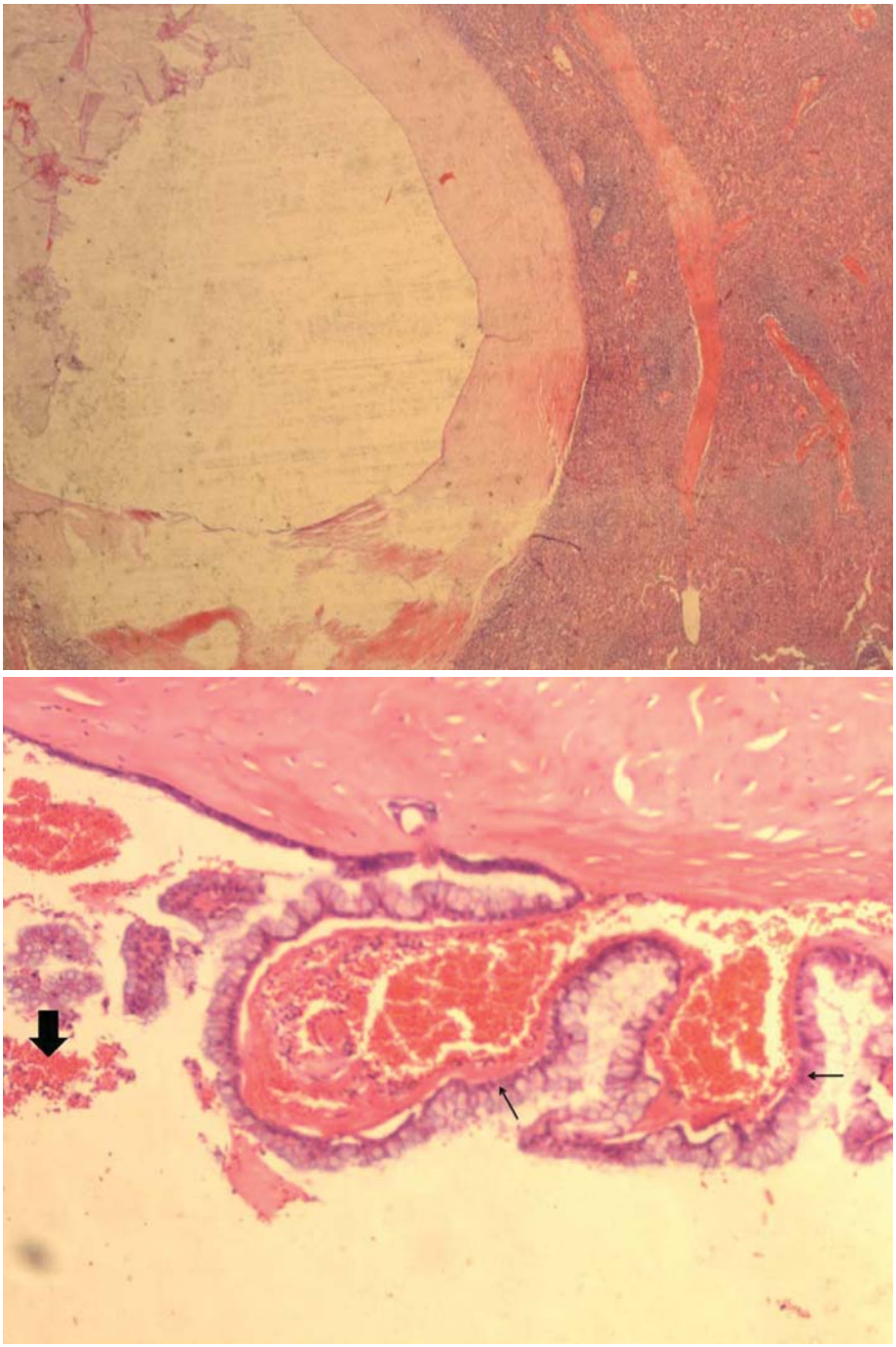


Figure 2. Cut surface of the cystic lesions showed a smooth inner surface with a gelatinous material.

these cysts had internal septa and peripheral calcifications. However, the liver, pancreas, and appendix were normal. Laboratory tests were unremarkable and splenectomy was performed on the 6th of December 2016.

Gross pathological examination showed an enlarged spleen covered with multiple variable sizes of cysts containing gelatinous material (Figure 1).

In addition, the cut surface of the cystic lesions has a smooth inner surface (Figure 2). Microscopic examination showed multiple cystic spaces that were lined by a single layer of columnar mucin-containing epithelium, without evidence of malignancy. The cystic spaces also contained mucin. The specimen contained no pancreatic tissue (Figures 3 and 4).



Figures 3 and 4. A mucin-containing cyst covered with a single layer of mucin-secreting columnar epithelium (thin arrows). The cystic cavity contains some mucin (thick arrow) (H&E).

The postoperative course was unremarkable and she was discharged fine.

DISCUSSION

Primary splenic cysts are rare lesions that are often discovered incidentally during surgeries for other reasons. However, the first case was reported in 1929.[8] A classification by Morgenstern et al. categorized splenic tumors into four: lymphoid, non-lymphoid, metastatic tumors, and tumor-like lesions including cysts and hamartomas. Vascular tumors are the most common type of non-lymphoid tumors. However, primary epithelial splenic tumors are extremely rare.[3] Martin classified splenic cysts according to the presence or absence of epithelial lining, type 1 cyst (true epithelial cysts), and type 2 cysts (false cysts without lining epithelium).[9]

Splenic mucinous cysts are cystic spaces that are lined by mucin-producing epithelium and that range from benign cystadenoma to malignant cystadenocarcinoma. The exact pathogenesis of splenic mucinous cysts is not fully understood, except for those arising from ectopic pancreas.[3] Moreover, ectopic pancreas in the spleen is a rare unusual finding.[10] Other related conditions include pseudomyxoma peritonei, mucocele of the appendix, and invagination of the splenic capsular epithelium into spleen.[3–7] Mucinous cystadenoma is also reported in other sites such as the ovary, pancreas, appendix, retroperitoneum, Fallopian tubes, lung, urinary bladder, and terminal ileum.[1]

The clinical presentation of patients with mucinous cystic tumors is nonspecific, but it depends on the site of the tumor.[1] Imaging by ultrasound, CT scan, or magnetic resonant imaging scan may reveal cystic lesions that may show internal septation or not. Peripheral calcification may be seen more frequently in the case of malignant cystadenocarcinoma than in benign cystadenoma; however, the scenario was not seen our case.[1] The definitive diagnosis is made only through histopathological examination.[1]

The limitation of this study is that the results cannot be generalized; therefore, more cases should be reported in order to have a better understanding about this very rare condition.

CONCLUSION

In our case, the diagnosis was primary mucinous cystadenoma of the spleen based on gross picture and histopathological examination of the splenic specimen. Mucinous cystadenoma of the spleen should be included in the differential diagnosis of patients presented with splenic lesions.

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