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MUCINOUS CYSTADENOMA OF THE SPLEEN IN SUDAN: REPORT OF A RARE CASE & REVIEW OF THE LITERATURE

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Abstract: A rare case of mucinous cystadenoma of the spleen is reported. It is the first case to be reported in Sudan and probably the third worldwide. The patient was a 33-year-old female with a multicystic spleen demonstrated by ultrasonography and CT-scan. Splenectomy was done. Histopathology and microscopy showed that the cystic space was lined by flattened & cuboidal epitheliod cells containing mucous material and surrounded by lymphoid follicles with no evidence of malignancy. English literature was reviewed and this case might be the third case of primary mucinous cystadenoma of the spleen in the world and the first in Sudan to be reported (Taiwanese and Japanese). Key words: spleen, mucinous cystadenoma, multicystic

Introduction and literature review

Mucinous cystadenomas are relatively uncommon benign cystic tumours[1]. The vast majority of them are found in the ovary, pancreas and appendix[1]. They have been identified in other unusual sites like retroperitoneum, Fallopian tube, lung, urinary bladder, liver[2], spleen[3] and terminal ileum[4]. The exact heterogenetic mechanism of splenic mucinous cystic tumours is unknown except those arising from heterotropic pancreatic tissue, or associated with pseudomyxoma peritonei[6-9] or with the mucocele of the appendix[5]. Also they are considered as invaginated splenic capsule mesothelium[5] although primary splenic cystadenomas are reported[5,10]. Splenic tumours are classified into four categories: lymphoid, non-lymphoid, metastatic and tumour-like lesions (cystic and hamartomatous)[6]. A tumour-like mimicking mucinous (colloid) cystadenocarcinoma in heterotropic pancreas of the prepyloric antrum were reported in the literature[10]. English literature was reviewed and only two cases of primary splenic mucinous cystadenoma were reported; one 63-year-old Taiwanese[11] and a 65-year-old Japanese man[3]. Three cases of splenic mucinous cystadenocarcinomas were reported[11,12]. In this paper a 33-year-old Sudanese female is reported with primary splenic mucinous cystadenoma.

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Case report

A 33-year-old Sudanese female was admitted to the Medical & Health Services Centre of the University of Khartoum on the 19th of January 2010 with multicystic splenomegaly. She was known to have antiphospholipid syndrome, history of seven abortions, one stillbirth & one neonatal death. She has 2 living daughters (age 10 & 8 years). She has history of appendecectomy in 1992 with no mucinous cystadenoma of the appendix. On physical examination she was well, 170cms height and 82kgs weight. Spleen was palpable 6cms below the costal margin. Ultrasonography and CT-scan showed multicystic spleen with no ascites or other abnormal findings. Laboratory tests were unremarkable. Splenectomy was done on the 20th of January 2010. The spleen was in form of multiloculated cystic mass with no communication to other organs. Appendix was removed with normal bed, normal ovaries, pancreas and peritoneum and no ascites. Gross pathological examination revealed a spleen measuring 14.5×10×5 cm & weighing 600 gm. The outer and inner surfaces of the spleen were covered by variably sized cysts, some containing gelatinous and yellowish fluid. Microscopy reported that the sections of the spleen showed multiple cystic spaces lined by flattened and cuboidal epithelial cells some of which contained mucinous material. These cystic spaces also contained mucin & were surrounded by lymphoid follicles. The red pulp was unremarkable. The diagnosis was mucinous cystadenoma of the spleen. The patient had unremarkable intra- & post-operative course.

Discussion

Mucinous cystadenoma of the spleen is a rare tumour. Its aetiology is not well understood & the exact heterogenetic mechanism of the splenic mucinous cystic tumours is unknown except those arising from heterotropic pancreatic tissue or associated with pseudomyxoma peritonii[6-9] or with mucocele of the appendix[5]. However they are considered as invaginated splenic capsule mesothelium[5]. Cystadenoma of the spleen is defined as cystic spaces lined by mucin-producing columnar cells[1]. Although mucinous cystadenoma is reported in the ovary, pancreas and appendix, it can occur in other unusual sites like retroperitoneum, Fallopian tubes, lung, urinary bladder, spleen & terminal ileum[2-4]. Few cases of the spleen were reported ranging from benign cystadenoma to a frankly malignant cystadenocarcinoma[11,12]. Splenic tumours are classified into four categories: lymphoid, non-lymphoid, metastatic & tumour-like lesion (cystic or hamartomatous)[6].

Our patient was a 33-year-old Sudanese female while two of the few reported cases in the literature were older; a 63-year-old Taiwanese woman[1] and a 65-year-old Japanese man[3]. The

mucinous cystadenoma of the spleen in our case was primary where no communication was identified with any other organ. Her appendix was removed in 1992 and no changes were identified in the spleen at that time with no cystadenoma of the appendex. She had a bad obstetric history having seven abortions, one still birth and one neonatal death. She was diagnosed to have antiphospholipid syndrome & whether it has something to do with mucinous cystadenoma of the spleen is not known.

In conclusion our case was diagnosed as primary mucinous cystadenoma of the spleen based on growth features (figure 1) & histopathology report of the spleen with the outer and inner surfaces covered by variable sized cysts, some containing gelatinous or yellowish fluid. Sections of the spleen showed multiple cystic spaces lined by flattened and cuboidal epithelial cells some of which contained mucous & were surrounded by lymphoid follicles. The red pulp of the spleen was unremarkable (figures 2 & 3). So mucinous cystadenoma of the spleen should be suspected in any patient with multiple cysts on spleen.

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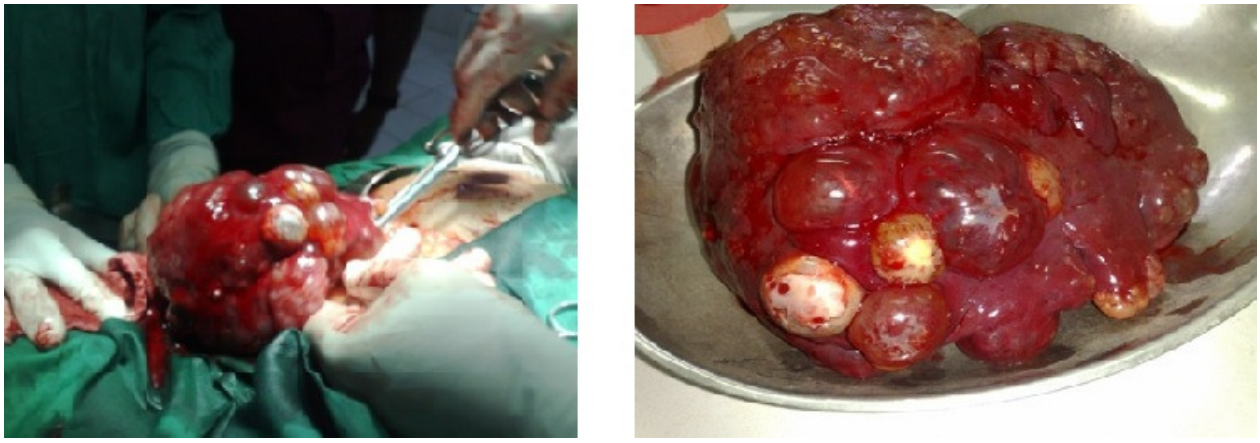

Figure 1: a. the appearance of the spleen intraoperatively & b. after removal

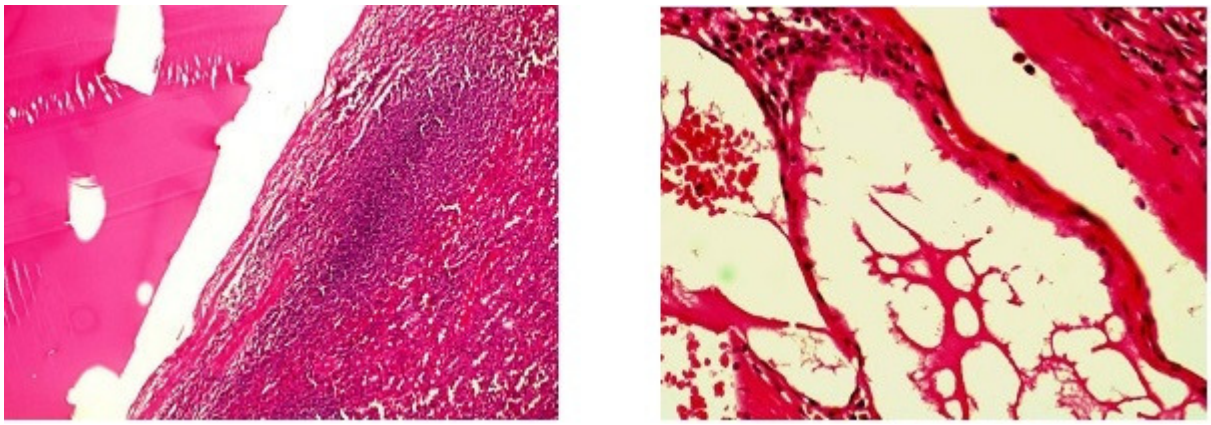

Figure 2: a. Mucin containing cyst surrounded by a nodule of white pulp and red pulp The cyst has no cellular line due to pressure atrophy (H&Ex40). b There are several cysts. Some contain mucin. Others contain red cells. The large mucin containing cyst in the center is lined in one part by cells with pale cytoplasm (H&Ex40)

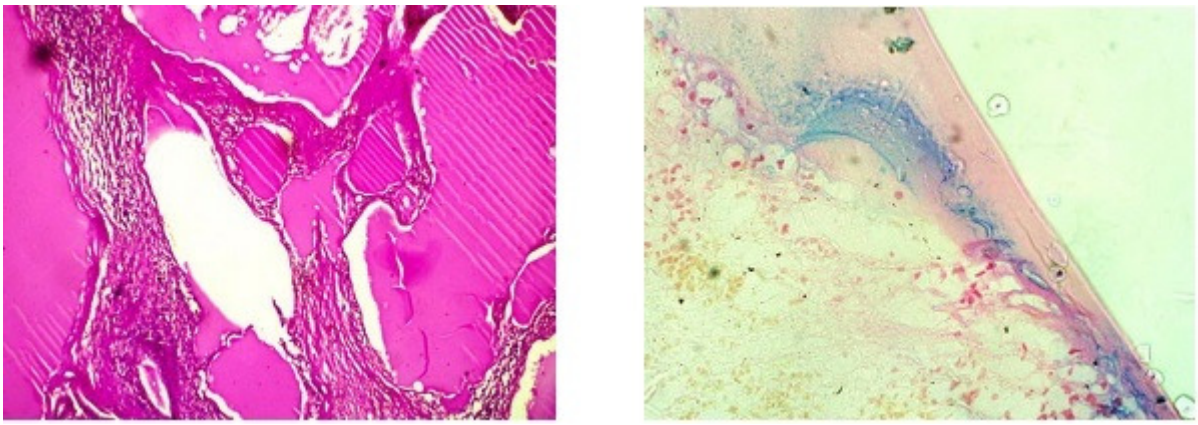

Figure 3: c Cysts containing PAS positive mucin (PASx40). d In very few cysts the mucin was positive for Alcian blue (Alcian blue stain x40)