

A Rare Case of Splenic Benign Mucinous Cystadenoma

Raafat A Hegazy¹, Abdelmonem A Hegazy², Salah F Alsayed³, Elsayed E Ammar⁴

¹Pathology Department, Faculty of Medicine, Zagazig University, Zagazig 44519, Egypt.

²Anatomy and Embryology Department, Faculty of Medicine, Zagazig University, Zagazig 44519, Egypt.

³Internal Medicine Department, Faculty of Medicine, Zagazig University, Zagazig 44519, Egypt.

⁴General Surgery Department, Kaleemullah Hospital, San-Alhagar, Zagazig 44519, Egypt.

Date of Submission: 27-06-2016

Date of Acceptance: 05-11-2016

Date of Publishing: 20-12-2016

ABSTRACT

This is a report of a rare case of benign mucinous cystadenoma of the spleen. The case was preceded by a benign mucinous cystadenoma of the ovary 10 years before, accompanied by pseudomyxoma peritonei. The pancreas and peritoneum were free. The spleen showed multilocular cystic mass with smooth inner surface and profuse inspissated mucus. Microscopically, no papillary structure was found on the inner surface of cystic space which was lined by a single layer of mucin-producing epithelial cells without malignant change. The cause of such rare case remains unclear. It is suggested that it may be a part of syndrome of multiple mucin producing cysts, like that of mucoviscidosis.

Keywords: Spleen, Cystadenoma, Mucinous, Benign.

INTRODUCTION

Tumors of the spleen are uncommon lesions. They are classified into two main categories; lymphoid and non-lymphoid tumors. The non-lymphoid tumors include a wide variety of lesions such as lipomas and pseudocysts. True epithelial cysts of the spleen are extremely rare^[1]. Only few cases of mucinous cystadenoma of spleen have been documented in the literature^[2-7]. Benign mucinous cystadenoma are relatively uncommon tumors. Most of them are found in the ovary, pancreas and appendix. However, they have also been identified in other unusual sites such as retroperitoneum, fallopian tube, lung, urinary bladder and liver^[8].

Name & Address of Corresponding Author

Dr. Abdelmonem Hegazy
Professor,
Department of Anatomy and Embryology
Faculty of Medicine, Zagazig University, Zagazig, 44519,
Egypt.
E mail: dr.abdelmonemhegazy@yahoo.com

In this article, we reported an extremely rare case of benign mucinous cystadenoma of spleen.

CASE REPORT

This is a female patient aged 65 years. Ten years ago, she was exposed to laparotomy because of huge uterus and cystic ovaries. The pelvis was full of mucin; and panhysterectomy was done. Histopathology showed benign mucinous cystadenoma of the ovary and pseudomyxoma peritonei. In the same year, she was presented with intestinal obstruction; resection anastomosis was done because of stenosis of mid-ileum. Histopathology showed fibrosis and inflammatory lesion in the ileum in addition to the pseudomyxoma peritonei.

Since the last year, she was complaining of fullness of epigastrium. Physical examination revealed a well-defined ovoid, non-tender, non-pulsatile and smooth surfaced mass that involved the left hypochondrium, epigastrium and umbilical regions, and was moving

with respiration. Pelvi-abdominal ultrasonography (US) was done. US revealed a cystic mass with no pelvi-abdominal collections. The mass was measuring 17 x 16 cm, extending from the epigastrium to the left side, with multiple loculations. Then, the case was followed by CT abdomen post IV contrast examination that showed a huge multicystic splenic lesion 165x110 mm, with turbid fluid [Figure 1,2]. Hydatid disease was suspected. However, hydatid antibodies' titer was investigated and appeared to be negative for the disease. Then, abdominal exploration was performed. Mid-line upper abdominal incision was done; and because of the large mass subcostal extension was done to prevent spillage of the mass. On exploratory laparotomy, a large tense and cystic mass arising from the diaphragmatic surface of spleen and involving the whole left hypochondrium with extension into the umbilical region and epigastric region displacing the stomach to right side, was found. Other common sites for cystic lesion such as pancreas, appendix, mesentery and liver were found normal. Splenectomy was done.

Gross pathological examination of the spleen showed a large cystic mass of 20x16x16cm in size and dark pink in colour with multiple yellowish cystic swellings [Figure 3]. The cut section showed multilocular cystic mass with smooth inner surface and profuse inspissated mucus [Figure 4]. Microscopically, no papillary structure was found on the inner surface of cystic space which was lined by a single layer of mucin-producing epithelial cells without malignant change [Figure 5-6]. The splenic tissue surrounding the cystic wall was of normal morphology. Histopathological examination was suggestive of benign mucinous cystadenoma of spleen. No pancreatic tissue was found in the specimen.

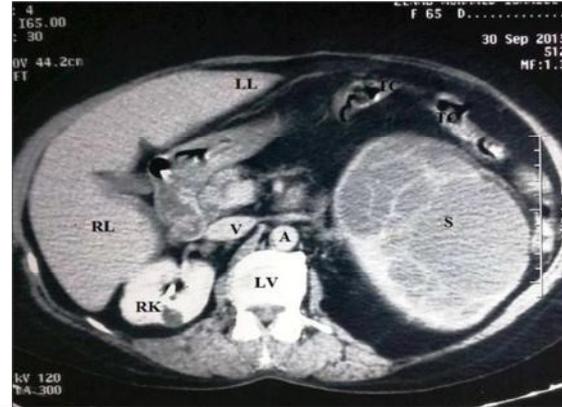


Figure 2: Transverse CT lower abdomen post IV contrast showing multilocular enlarged spleen (S) displacing the transverse colon (TC) forwards. Left (LL) and right lobes (RL) of liver, right kidney (RK), inferior vena cava (V), aorta (A) and body of lumbar vertebra (TV) are identified.



Figure 3: A photograph of the enlarged spleen showing its gross picture with multiple yellowish bulging cysts and irregular outline.



Figure 1: Transverse CT upper abdomen post IV contrast showing multilocular enlarged spleen (S) displacing the fundus of the stomach (F) towards the left lobe of liver (LL). Body of thoracic vertebra (TV) and aorta (A) appear.



Figure 4: A photograph of the spleen showing cut section of the spleen with multilocular cystic lesion. The cysts (C) are of different sizes with smooth inner surface.

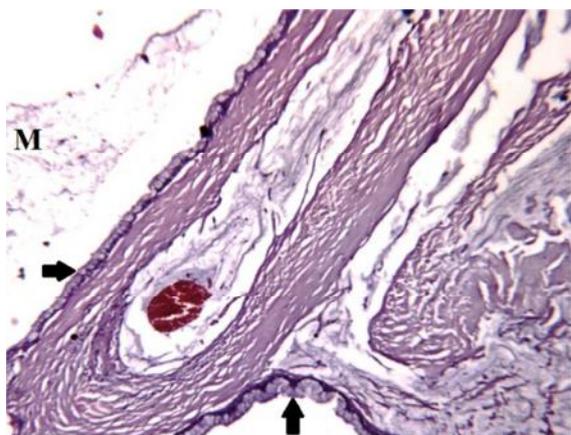


Figure 5: A microscopic picture of splenic cysts lined with simple epithelium (arrows) and containing mucin (M). A blood capillary is noticed in the stroma. X150.

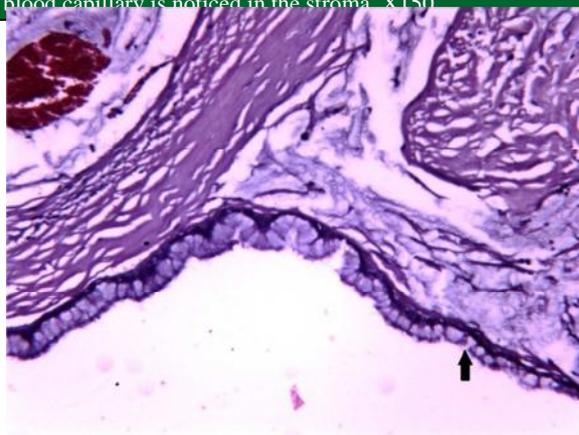


Figure 6: A higher magnification of the previous figure showing a single layer of cells (arrow) lining the cyst. The cells are tall columnar with supranuclear mucin vacuoles. No atypia of the cells is noticed. X400.

DISCUSSION

Benign mucinous cystadenoma of spleen is an extremely rare finding [2-4,7]. Benign mucinous cystadenoma in other organs such as ovary, pancreas and appendix are relatively uncommon [8]. In our case, the patient suffered from benign mucinous cystadenoma of ovary 10 years ago. The case was associated with pseudomyxoma peritonei. Pseudomyxoma peritonei recurred in the same year. The pancreas and appendix were free. The origin of cystadenoma of spleen is either primary or secondary. Metastatic mucinous cystadeno carcinoma from the ovary to the spleen is a possibility. However, metastatic tumors of the spleen are very rare despite it is one of the most vascular organs in the body [9]. Explanations proposed for the relative paucity of

splenic metastases have included the sharp angle made by the splenic artery which makes it difficult for tumour emboli to enter the spleen; the rhythmic contractile nature of the spleen which squeezes out the tumour emboli; the absence of afferent lymphatics to carry metastatic tumour to the spleen; and antitumour activity due to a high concentration of lymphoid tissue in the spleen [9]. Another possibility is that the tumor occurred in the ovary 10 years ago was malignant and that discovered now in the spleen is well-differentiated malignant tumor. But the long period of history (more than 10 years), the less liability of metastasis in the spleen and the absence of other metastasis in the body of the patient who did not receive any anti-cancer treatment indicate no malignancy. Moreover, the lesion of the spleen in the current case was completely benign as shown by histopathological examination. All these notices exclude the possibility of malignant mucinous cystadeno carcinoma.

The report of CT scan gave a possibility of hydatid cyst of the spleen; against this opinion was the eosinophils in blood was (4%). Moreover, the gross and histopathological picture ruled out this possibility. The question arises in this situation is about the origin of benign cystadenoma in the spleen; whether invagination of the splenic capsule [3], or the presence of heterotopic remnants of pancreatic tissue in the ovary, and spleen in the present case [10]. It is suggested that this case might be a part of syndrome of multiple mucin producing cysts, like that of mucoviscidosis.

CONCLUSION

This is report of a case of benign mucinous cystadenoma of the spleen. More investigations and studies are recommended to clarify and explain such rare cases.

Acknowledgments

No external funding was provided.

Compliance with ethics guidelines

The authors declares that he has no conflict of interest. This study was done after approval by the relevant institutional review board or ethics committee and consent of the patient.

REFERENCES

1. Morgenstern L, Rosenberg J, Geller SA. Tumors of the spleen. *World J Surg.* 1985; 9(3): 468-476.
2. Kruslin B. Pseudomyxoma peritonei associated with mucinous epithelial cysts of the spleen. *Lijec Vjesn.* 2001;123(5-6):154.
3. Morinaga S, Ohyama R, Koizumi J. Low-grade mucinous cystadenocarcinoma in the spleen. *Am J Surg Pathol.* 1992; 16(9): 903-908.
4. Nisar PJ, Zaitoun AM, Lobo DN, Rowlands BJ. Heterotopic pancreas in the spleen: malignant degeneration to mucinous cystadenocarcinoma. *Eur J Gastroenterol Hepatol.* 2002; 14(7): 793-796.
5. Dedic N, Premuzic M, Cavka S, Ostojic R, Hrstic I, Vucelic B. Pseudomyxoma peritonei associated with splenic mucinous epithelial cysts--case report. *Lijec Vjesn.* 2000; 122(11-12):272-275.
6. Kapoor S, Naik S, Sharma S, Varshney S. Pseudomyxoma peritonei due to a ruptured mucinous cystadenoma of the spleen. *European Surgery.* 2007; 39 (5):314-316.
7. Hirota M, Hayashi N, Tomioka T, Murakami S, Ohshima H, Yamasaki K, Miyamoto J, Tamiya S, Ogawa M. Mucinous cystadenocarcinoma of the spleen presenting a point mutation of the Kirsten-ras oncogene at codon 12. *Dig Dis Sci.* 1999; 44(4): 768-774.
8. Singh O, Gupta S, Shukla S, Mathur RK: Case report: A rare case of primary mucinous cystadenoma of spleen. *J Clin Med Res.* 2009;1(4):237-239.
9. Giovagnoni A, Giorgi C, and Goteri G: Tumours of the spleen. *Cancer Imaging.* 2005; 5(1): 73-77
10. Zanetti G, Riccioni L, Gallo C, Salfi N, Martinelli GN. Splenic mucinous cystadenocarcinoma arising in heterotopic pancreatic tissue. *Tumori.* 1998;84(5):606-610.

Copyright: Academia Anatomica International is an Official Publication of "Society for Health Care & Research Development". This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

How to cite this article: Hegazy RA, Hegazy AA, Alsayed SF, Ammar EE. A Rare Case of Splenic Benign Mucinous Cystadenoma. *Acad. Anat. Int.* 2016;2(2):7-10.

Source of Support: Nil, **Conflict of Interest:** None declared.