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Case Report

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Sclerosing Angiomatoid Nodular Transformation Disguised as Splenic Metastasis: A Case Report

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Abstract

Background: SANT (Sclerosing Angiomatoid Nodular Transformation) is a relatively rare, benign vascular lesion of the spleen, usually found incidentally during un-intended medical procedure. Most of patients with SANT have no clinical symptoms and difficult to make differential diagnosis with any imaging modalities. Definite diagnosis needs to be confirmed pathologically, post-operatively.

Case presentation: This study reports a rare case of SANT in a 62-year-old female patient who underwent sigmoid colectomy eleven years ago due to stage II colon cancer. Splenic mass was found by computed tomography (CT) of the abdomen. Radiologist suggest to rule-out metastatic mass or other primary splenic malignancy. The patient underwent laparoscopic splenectomy and SANT was finally diagnosed with pathologic findings.

Conclusion: SANT of spleen can also disguise as splenic metastasis, surgical resection seems to be necessary not only for definite diagnosis of SANT, but also relieving patient's anxiety from metastatic disease.

Keywords: Splenic metastasis; Sclerosing angiomatoid nodular transformation; Spleen.

Introduction

Spleen is sometimes referred to as the "Neglected organ" because it is always seen on computed tomography (CT) or Magnetic Resonance Imaging (MRI) scans, but rarely develops disease [1]. In addition, splenic metastases from solid tumors are considered an exceptional clinical diagnosis and are usually found as part of a broader metastatic disease. Sclerosing angiomatoid nodular transformation (SANT) of the spleen, first reported in the literature by Martel et al. [2]. in 2004, is a rare benign non-neoplastic vascular splenic lesion of uncertain etiology that often masquerades as a potentially malignant entity during the diagnostic process, but surgery remains the only method of diagnosis. In this paper we report a case of SANT of the spleen operated at our hospital.

Case report

A 67-year-old female with appropriately managed and monitored AJCC stage II sigmoid colon cancer surgery on 2006, was newly found to have a splenic incidentaloma on CT of the abdomen and pelvis on 2017. The patient had no previously diagnosed

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medical conditions other than colon cancer and no family history of cancer. The patient was treated with six cycles of leucovorin/5-fluorouracil after colon cancer surgery. The patient suffered from abdominal discomfort approximately two months ago to this visit and underwent colonoscopy and CT scan. All of her blood tests, including CEA (carcinoembryonic antigen), were normal. The abdomen was unremarkable except for a low midline laparotomy scar from a previous open anterior resection for colon cancer. Colonoscopy and CT revealed no sign of local and systemic tumor recurrence. However, CT scan with contrast in late arterial phase demonstrated a pole of spleen (Figure 1). The radiological differential diagnosis was focused on suspected metastatic mass or primary splenic angiosarcoma. Based on this CT finding, the patient underwent a laparoscopic splenectomy under the diagnosis of metastatic splenic tumor. This tumor was finally proved by pathological examination to be a SANT of spleen. Macroscopically, the spleen, measuring 13.0x6.0x5.0 cm, was enlarged by the mass, which measured 6.0x5.0 cm. The cut surface of the splenic mass was firm, gritty consistency with variegated appearance (Figure 2A). Microscopically, the mass was composed of multiple vascular structures separated by fibrous connective tissue (Figure 2B and C) and immunohistochemical analysis revealed positive staining for CD31, CD34 (Figure 3A and B) and negative for CD8 (Figure 3C) consistent with diagnosis of a cord capillary-like type of the SANT. Recovery was satisfactory and the patient was discharged on the 14th postoperative day with no complications.

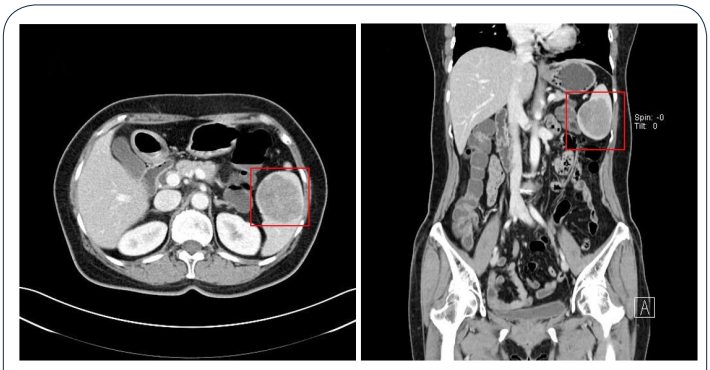

Figure 1: Late arterial phase contrast enhanced computed tomography of the abdomen and pelvis revealed a 5.4x4.5 cm well-circumscribed, heterogeneously enhancing solid splenic mass (red rectangles) in axial **(A)** and coronal **(B)** planes.

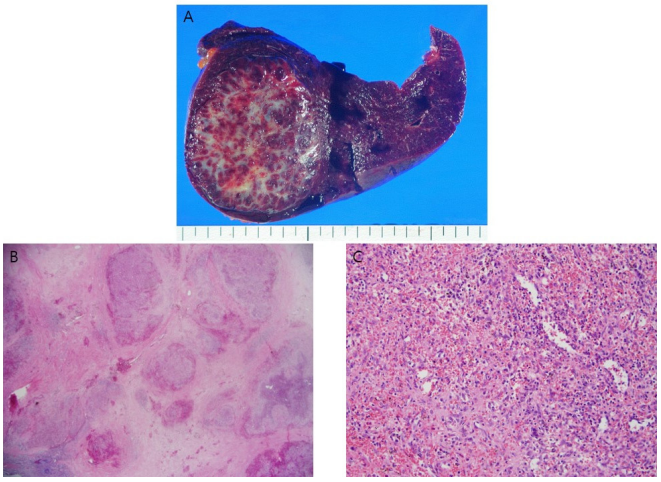

Figure 2: The cut surface of the spleen showed well defined round mass with multiple red tan nodules surrounded by whitish fibrotic tissue **(A)**. The center of nodule showed multiple small blood vessels with loose stroma surrounded by fibrous tissue. H&E, x12.5 **(B)**, x200 **(C)**.

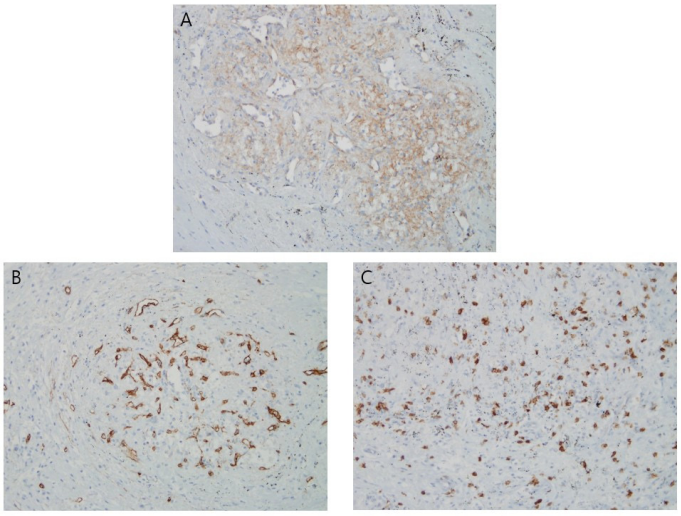

Figure 3: Immunohistochemical staining of the small blood vessels within the lesion were positive for both CD31 **(A)** and CD34 **(B)** and negative for CD8 **(C)**. H&E, 200.

Discussion

SANT of the spleen is rare, newly recognized, non-neoplastic vascular lesion of the spleen whose pathogenesis is still uncertain. It usually found in middle-aged adults, slightly female dominant but gender bias seems to be vanished and neutralized as more cases are collected [4]. Most of patients have no clinical symptoms, diagnosed by incidentally during imaging or other medical procedure of un-related purpose. For symptomatic cases, abdominal pain or discomfort is predominant.

In gross appearance of SANT is solid, well-circumscribed, non-encapsulated nodular lesion distinct from splenic parenchymal. The pathologic features of SANT is creating the angiomatoid nodules mixture of spindle cells, inflammatory infiltrate, and endothelial vascular proliferations [5]. SANT has three district types of immunophenotype compared to normal composition of splenic red pulp [6]. The first one is cord capillary-like type, consists of well-formed cord capillaries in an organized lobular arrangement in lining endothelial cells, expresses CD34 and CD31 but not for CD8. The second type, vessel consistent with splenic sinusoids and include endothelial cells expresses CD31, CD8 but not for CD34. The third type consists of small veins are arranged complex mesh like patterns expresses only CD8, not for CD34 and CD31. In our case, it was compatible cord capillary-like type [7].

The differential diagnosis of splenic mass is various and there are many benign and malignant lesions. Cystic lesion of the spleen; congenital true cysts, post-traumatic pseudocyst, etc; can be identify by imaging modalities with ease. Solid lesion of the spleen however, have more difficulty in differential, such as hemangioma, hamartoma and angiosarcoma, etc. Hemangiomas of the spleen can be distinguished from SANT by MRI scan by their high T2 signal intensity. Hamartomas and angiosarcoma also can be seen hyper-intensity on T2-weighted images, distinguished from SANT.

Rachel BL et al. [8]. report characteristic CT and MRI findings of SANT, including peripheral enhancing radiating lines in arterial or portal phase, progressive enhancement, hypo-intensity on T2-weighted image, CT scan shows "spoke wheel" pattern, the result of contrast penetrating the center of the mass from the vascular rim, progressive central enhancement with delayed imaging. Although there are some notable findings suggest SANT, definite diagnosis still impossible without tissue diagnosis, especially in patients who diagnosis malignant disease previously.

SANT of the spleen usually found as incidentaloma, and difficult to diagnosed just by imaging modalities, surgical resection seems to be necessary eventually. SANT considered as benign, primary non-neoplastic lesion, Weinreb et al. [9]. report that core needle biopsy can be used diagnostic option for tissue confirm. However there is important issue which the risk of intraperitoneal seeding if the lesion proved by malignant feature, such as metastatic tumors or angiosarcoma.[4] Ruper L et al [10]. report a case of rapidly growing SANT for 1 year follow-up who underwent anterior resection for stage III rectal cancer. Due to including malignant pathologic disease in differential diagnosis for SANT, and there is no reliable radiologic feature has been identified to distinguishing between these conditions currently, SANT would be diagnosed on the basis of surgical histopathology for a while.

Conclusion

Most of the patients with pathologically confirmed SANT of the spleen were asymptomatic preoperatively and thus were usually found incidentally as a splenic incidentaloma prior to surgery [11]. Unfortunately, there are no definitive diagnostic imaging methods that can differentiate between SANT and splenic metastases before surgery without pathologic confirmation [12]. Most of the cancer survivors who incidentally detected splenic mass prefer to undergo splenectomy due to fear of metastasis. Therefore, to avoid unnecessary psychological stress or splenectomy, we believe that efforts should be made to perform relatively non-invasive diagnostic methods such as preoperative image-guided splenic core biopsy. Further research should focus on clinical and radiological diagnosis of SANT as well as on treatment of patients with asymptomatic and small findings.

Declarations

Ethics approval and consent to participate: Institutional Ethical committee approved the study. Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

Consent for publication: Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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All authors read and approved the final manuscript.

References

1. Giovagnoni A, Giorgi C, Goteri G. Tumours of the spleen. Cancer Imaging. 2005;5:73-77.
2. Martel M, Cheuk W, Lombardi L, Lifschitz-Mercer B, Chan JK, Rosai J. Sclerosing angiomatoid nodular transformation (SANT): Report of 25 cases of a distinctive benign splenic lesion. Am J Surg Pathol. 2004; 28:1268-1279.
3. Bamboat ZM, Masiakos PT. Sclerosing angiomatoid nodular transformation of the spleen in an adolescent with chronic abdominal pain. J Pediatr Surg. 2010; 45: E13-16.
4. Falk GA, Nooli NP, Morris-Stiff G, Plesec TP, Rosenblatt S. Sclerosing Angiomatoid Nodular Transformation (SANT) of the spleen: Case report and review of the literature. Int J Surg Case Rep. 2012; 3: 492-500.
5. El Demellawy D, Nasr A, Alowami S. Sclerosing angiomatoid nodular transformation of the spleen: case report. Pathol Res Pract. 2009; 205: 289-293.
6. Pradhan D, Mohanty SK. Sclerosing angiomatoid nodular transformation of the spleen. Arch Pathol Lab Med. 2013; 137: 1309-1312.
7. Kim HH, Hur YH, Koh YS, et al. Sclerosing angiomatoid nodular transformation of the spleen related to IgG4-associated disease: report of a case. Surg Today. 2013; 43: 930-936.
8. Lewis RB, Lattin GE, Jr., Nandedkar M, Aguilera NS. Sclerosing angiomatoid nodular transformation of the spleen: CT and MRI features with pathologic correlation. AJR Am J Roentgenol. 2013; 200: W353-360.
9. Weinreb I, Bailey D, Battaglia D, Kennedy M, Perez-Ordonez B. CD30 and Epstein-Barr virus RNA expression in sclerosing angiomatoid nodular transformation of spleen. Virchows Arch. 2007; 451: 73-79.
10. Langer R, Dinges J, Dobritz M, et al. Sclerosing angiomatoid nodular transformation of the spleen presenting as a rapidly growing tumour in a patient with rectal cancer. BMJ Case Rep. 2009; 2009.
11. Ong BS, Thomas R. Sclerosing Angiomatoid Nodular Transformation (SANT): A Rare Splenic Tumor and Unusual Cause of Anemia. Am J Case Rep. 2021; 22: e933598.
12. Efared B, Sidibe IS, Erregad F, Hammas N, Chbani L, El Fatemi H. Sclerosing angiomatoid nodular transformation of the spleen (SANT) in a patient with clear cell carcinoma of the uterus: A case report. J Med Case Rep. 2018; 12 :377.