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Scerosing angiomatoid nodular transformation of the spleen presenting rapid growth after adrenalectomy: Report of a case



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ABSTRACT

INTRODUCTION: Scerosing angiomatoid nodular transformation (SANT) is a rare benign lesion with an unknown natural history and pathogenesis. So far fewer than 100 cases were documented, but detailed incidence and prevalence are unknown.

PRESENTATION OF CASE: We report a case of SANT of the spleen in a 37-year-old man that showed rapid growth after adrenalectomy for primary aldosteronism. Computed tomography showed a nodule in the spleen that increased in size from 2.0 cm to 7.0 cm during 3 years of observation.

DISCUSSION: This case is reported because data regarding growth rates and natural history of these lesions are limited and few cases have been reported to show the rapid growth progression seen in this case.

CONCLUSION: Decreases in glucocorticoid concentrations following adrenalectomy may have contributed to the rapid growth of SANT of the spleen, because SANT is considered to be related to immunoglobulin G4-associated disease.

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1. Introduction

Scerosing angiomatoid nodular transformation (SANT) is a very rare benign lesion in the spleen, initially described in 2004 by Martel et al. [1]. So far fewer than 100 cases were documented, but detailed incidence and prevalence are unknown. SANT is composed of angiomatoid nodules in a fibrosclerotic background [2]. Histologically, the angiomatoid nodules comprise several types of blood vessels distinguished by their immunohistochemical profiles, including a cord capillary-like type (CD31+CD34+CD8-), small veins (CD31+CD34-CD8-), and a sinusoid-like type (CD31+CD34-CD8+) [1,3].

SANT appears to follow a benign clinical course, with splenectomy being curative, but data regarding the natural history and growth rates of this disease are scarce. The pathogenesis of SANT also remains unclear. SANT has been hypothesized to represent a peculiar hamartomatous transformation of splenic red pulp in response to exaggerated non-neoplastic stromal proliferation [3]. On the other hand, SANT is reportedly associated with Epstein-Barr virus (EBV) infection [2], and immunoglobulin (Ig)G4-related sclerosing disease [5,6]. We report herein a case of SANT of the spleen presenting rapid growth after adrenalectomy for primary aldosteronism. We chose splenectomy by hand assisted laparoscopic surgery (HALS), did not perform minimal invasive surgery such as

tumorectomy, because we could not completely deny a malignant lesion.

We discuss the possible pathogenesis and treatment of SANT.

2. Case report

A 33-year-old man was admitted for SANT. His medical history had laparoscopic adrenalectomy for primary aldosteronism. Physical examination of the abdomen showed no abnormalities. Abdominal computed tomography (CT) performed 1 year after surgery incidentally revealed a hypodense splenic lesion (20 mm) (Fig. 1). Physical examination of the abdomen showed no abnormalities. Laboratory examinations including serum carbohydrate antigen (CA)19-9 and carcinoembryonic antigen (CEA) concentrations were within normal limits. Plain CT showed a low-density, circular lesion with a well-circumscribed border. No cystic changes, necrosis or calcification was evident in the lesion. Contrast-enhanced CT showed delayed enhancement centripetally, with a wheel-like appearance. Abdominal magnetic resonance imaging (MRI) demonstrated a nodule in the spleen with signal hypointensity on T1-weighted imaging, and heterogeneous signal hyperintensity on T2-weighted imaging (Fig. 2).

The main radiological differential diagnoses were hamartomas, sclerosed hemangioma, and inflammatory pseudotumor. As no sign of malignancy was present, we decided to observe the progress of the lesion without intervening. Two years later, CT showed the mass was appeared to be progressive (50 mm). The next year, the mass had increased in size to 70 mm (Fig. 1). Because malignant

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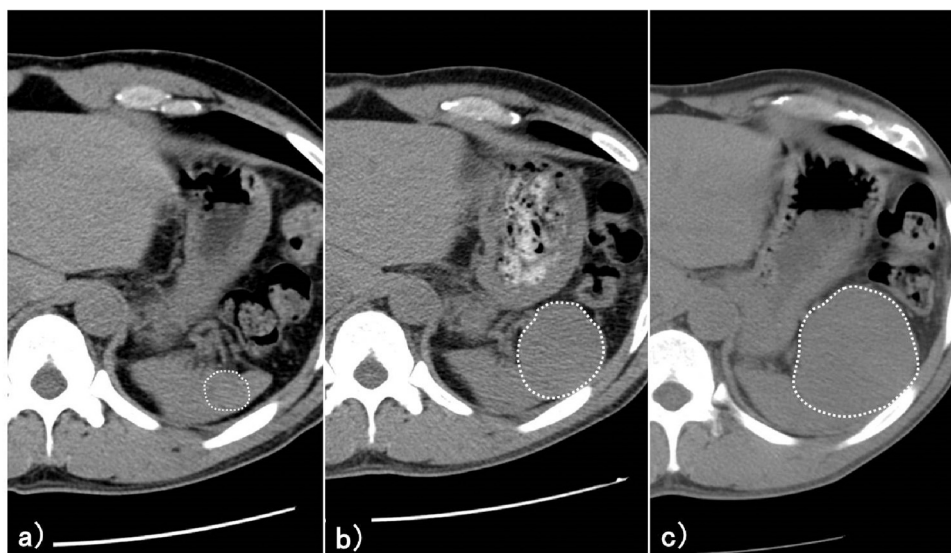


Fig. 1. Abdominal computed tomography (CT). a) A 2.0-cm mass is evident in the spleen 1 year after adrenalectomy. b) Three years after adrenalectomy, the mass had increased in size to 5 cm c) The next year, the mass has increased in size to 70 mm.

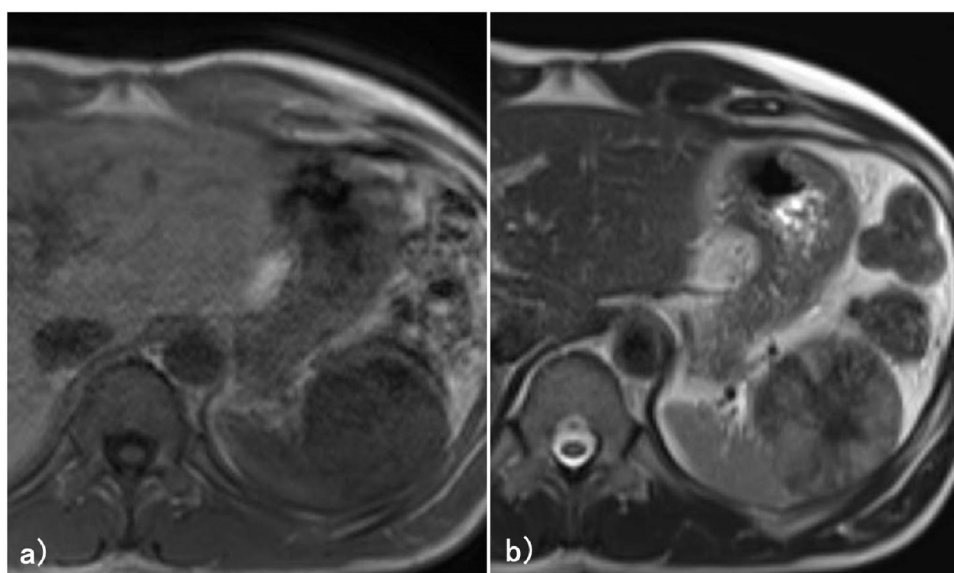


Fig. 2. Magnetic resonance imaging (MRI). a) A signal-hypointense nodule is seen in the spleen on T1-weighted imaging. b) Heterogeneous signal hyperintensity is seen on T2-weighted imaging.

neoplasm could not be completely excluded given such progressive growth, splenectomy was performed.

We chose hand assisted laparoscopic surgery (HALS) by lateral decubitus position, because the tumor located at the spleen and it was not invasive. (Fig. 3)

Gross examination revealed a well-demarcated, solitary lesion measuring 70 × 65 mm, containing a central bulky stellate mass of white compact tissue (Fig. 3). Histopathological findings showed three types of vascular structure in the tumor: capillary-like vessels; dilated sinusoid-like vessels; and ectatic veins. Neither nuclear atypia nor necrosis was present. Immunohistochemical analysis revealed a complex vascular pattern, in which the capillaries showed CD34+/CD8-/CD31+, the dilated sinusoid-like vessels showed CD34-/CD8+/CD31+, and small veins showed CD34-/CD8-/CD31+. In the sclerotic stroma, some plasma cells revealed a IgG4+ status (Fig. 4). EBV was not detected according to EBV-encoded RNA (EBER). The pathological diagnosis was SANT.

3. Discussion

SANT of the spleen is rare and mostly recognized as an incidental finding on routine medical imaging, as in this case. The patients are usually asymptomatic but sometimes an abdominal discomfort or pain can be observed. Zhixin et al. reviewed 127 patients with SANT. In this report, a slight female preponderance is evident, and predominantly affects middle-aged adults according to a review [7]. Although the exact nature of this disease remains unclear, SANT is considered benign and does not present with recurrence or malignant behavior [8]. In our case, the splenic lesion displayed a rapid growth in size from 20 mm to 70 mm during 3 years after adrenalectomy. Surprisingly, CT showed no lesion in the spleen just before adrenalectomy, and CT 1 year after adrenalectomy revealed a 20-mm lesion in the spleen. Although growth of SANT on follow-up imaging is reportedly common [9], few reports have shown such rapid growth as seen in the present case. Of course, these findings

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