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CASE REPORT

Sclerosing angiomatoid nodular transformation of the spleen, a rare cause for splenectomy: Two case reports

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Author contributions: Chikhladze S and Aumann K initiated the study and collected the cases; Wittel UA, Fichtner-Feigl S, and Chikhladze S performed the surgery; Werner M and Aumann K performed histological evaluations; Lederer AK wrote the manuscript with the help of Chikhladze S and Aumann K; Wittel UA, Fichtner-Feigl S, and Werner M revised the manuscript.

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Abstract

BACKGROUND

Sclerosing angiomatoid nodular transformation (SANT) is a rare benign disease of the spleen with unknown origin. Clinical symptoms are inhomogeneous, and suspicious splenic lesion often found incidentally, leading to splenectomy, as malignancy cannot securely be ruled out. Diagnosis is made histologically after resection.

CASE SUMMARY

Two cases of German, white, non-smoking, and non-drinking patients of normal weight are presented. The first one is a 26-year-old man without medical history who was exhibiting an undesired weight loss of 10 kg and recurring vomiting for about 18 mo. The second one is a 65-year-old woman with hypertension who had previously undergone gynecological surgery, suffering from a lasting feeling of abdominal fullness. Both showed radiologically an inhomogeneous splenic lesion leading to splenectomy approximately 6 and 9 wk after surgical presentation. Both diagnoses of SANT were made histologically. Follow-up went well, and both were treated according to the recommendation for asplenic patients.

CONCLUSION

SANT is a rare cause of splenectomy and an incidental histological finding. Further research should focus on clinical and radiological diagnosis of SANT as well as on treatment of patients with asymptomatic and small findings.

Key words: Splenectomy; Sclerosing angiomatoid nodular transformation; Spleen; Abdominal discomfort; Gastrointestinal dysfunction; Case report

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Core tip: We are presenting two cases of clinical evident sclerosing angiomatoid nodular transformation (SANT) and discuss the challenge of clinical non-operative management of small and asymptomatic splenic lesions. SANT is a benign vascular lesion of unknown etiology occurring in the spleen and does not appear to be related to age, gender, or pre-existing illnesses, although some reports have reported a higher frequency in women. SANT is an incidental histologically finding after splenectomy, and there is no guideline for treatment of SANT.

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INTRODUCTION

Abdominal discomfort and gastrointestinal dysfunction are common reasons for a doctor's consultation. Nearly a third of acute cases appear to be nonspecific without any organic lesion being causative for the complaint[1]. Long-lasting symptoms lead to a variety of diagnostic procedures, which in turn often lead to incidental findings[2]. A really rare incidental finding is sclerosing angiomatoid nodular transformation (SANT) of the spleen. SANT is a benign vascular lesion of unknown etiology occurring in the spleen and was first described in 2004[3]. SANT does not appear to be related to age, gender, or pre-existing illnesses, although some reports have reported a higher frequency in women[4]. Some patients with SANT complain about abdominal discomfort, which often includes nausea and vomiting, as well as nausea, vomiting, and occasional malnutrition[3,5,6]. As a vascular and splenic tumor, SANT might lead to anemia or anemia-related fatigue[3,7,8]. Contrarily, some patients can be asymptomatic[3-5,9,10]. SANT is often clinically and radiologically confused with hemangioma or hamartoma[9] and can only be proven histologically[3,8]. Because of the supposed increased risk of a spontaneous rupture of large vascular splenic lesions and the risk for a possible malignancy of the suspicious lesion, splenectomy should be performed[11]. Recent publications have focused on pathologic findings, which is why we overview the clinical symptoms and surgical treatment of SANT with the help of two case reports.

CASE PRESENTATION

According to the CARE guidelines for publication of case reports[12], this report covers two patients with histologically proven SANT of the spleen who underwent splenectomy at the Department of General and Visceral Surgery of the University Medical Center of Freiburg, Germany.

Chief complaints

Case 1: A 26-year-old Caucasian, non-smoking, and non-drinking man (175 cm, 65 kg, body mass index: 21.2 kg/m^2) exhibiting an undesired weight loss of 10 kg and recurring vomiting for about 18 mo.

Case 2: A 65-year-old Caucasian, non-smoking, and non-drinking woman (150 cm, 50 kg) presented with a lasting feeling of abdominal fullness.

History of present illness

Case 1: Physical examination and initial gastroenterological evaluation as well as tests for food intolerances revealed no pathologic results. Intake of medication to alleviate symptoms was negated.

Case 2: Physical examination showed normal findings, and the initial gastroenterological evaluation revealed no pathologic results. Intake of medication to alleviate symptoms was negated.

History of past illness

Case 1: Nothing to declare.

Case 2: Patient suffered from hypertension (prescribed with once daily medication of candesartan 4 mg) and underwent previous gynecological surgery (curettage and polypectomy).

Personal and family history

Case 1: Nothing to declare.

Case 2: Family history showed one case of melanoma (father) and one case of breast cancer (sister).

Imaging examinations

Case 1: Abdomen magnetic resonance imaging showed a 46 mm × 41 mm × 44 mm heterogeneous solid mass of the spleen. According to the magnetic resonance imaging diagnosis, it was most likely a thrombosed hemangioma.

Case 2: Abdomen computed tomography showed a 74 mm × 54 mm heterogeneous solid mass of the spleen appearing to be an angiosarcoma rather than a hemangioma (Figure 1). Furthermore, a suspicious lesion of the pancreas, most likely an intraductal papillary mucinous neoplasm, was found.

FINAL DIAGNOSIS

Case 1

Grossly, the spleen was enlarged to 140 mm × 82 mm × 43 mm due to a solitary lesion, 41 mm × 37 mm × 32 mm, sharply demarcated and composed of red-brown nodules in dense fibrous stroma (Figure 2). The spleen weighed 260 g. Histologically, the lesion appeared micronodularly with slit-like and irregular shaped vascular spaces lined by inconspicuous endothelial cells. Stroma contained dense fibrous tissue with scattered myofibroblasts, inflammatory cells, and numerous red blood cells. The nodules were surrounded by dense collagen fibers. Increased immunoglobulin G4-positive plasma cell population was not found. Neither necrosis nor atypia was seen, and the mitotic rate was very low. By RNA *in situ* hybridization, Epstein-Barr virus was not detectable. The diagnosis of SANT was rendered.

Case 2

The spleen was 105 mm × 82 mm × 59 mm, widely fibrotic, and scarred, showing calcification and residuals of old bleedings in an intact capsule. The spleen weighed 198 g. Histologically, the lesion presented tumorous proliferation of partly nodular, partly lobular endothelial cells with diffuse lymphohistiocytic infiltration, distinct sclerosis, and extensive hystiocytic aggregates with displacement of local parenchyma. Increased immunoglobulin G4-positive plasma cell population was found. Neither necrosis nor atypia was seen, mitotic rate was very low. By RNA in situ hybridization, Epstein-Barr virus was not detectable. The diagnosis of SANT was made.

TREATMENT

Case 1

Surgical presentation led to the indication for laparoscopic splenectomy. The patient consented to surgical resection and was recommended to consult his family doctor before operation to be vaccinated against *Haemophilus influenza*, *Streptococcus pneumoniae*, and *Neisseria meningitidis*, according to the guideline for asplenic patients[13].

Operation was performed electively in general anesthesia approximately 6 wk after surgical presentation (Figure 3). Inspection of abdomen revealed no further pathologies. Patient showed an accessory spleen, which was not removed. Operation was performed as previously described by others[14,15]. After complete laparoscopic mobilization of the spleen, a 4 cm median laparotomy of the upper abdomen was performed to retrieve the organ in a retrieval bag. Operation lasted 106 min, and bleeding was minimal. Patient was taken to the post anesthesia care unit after operation. Patient did well following surgery and was discharged on the 4th postoperative day.

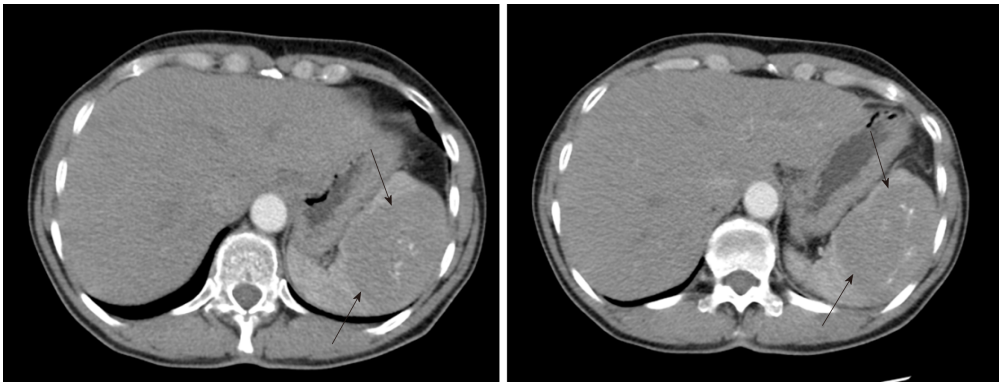

Figure 1 Abdomen computed tomography of a 65-year-old female patient showing a 74 mm × 54 mm solid splenic lesion (marked with arrows).

Case 2

Surgical presentation led to an indication for laparoscopic splenectomy. Patient was recommended to consult her family doctor for necessary vaccinations before operation.

Operation was performed electively under general anesthesia approximately 9 wk after surgical presentation (Figure 3). Inspection of abdomen revealed no further pathologies. As mentioned above, operation was performed as it was previously described by others[14,15]. After complete laparoscopic mobilization, the spleen was placed in a retrieval bag. For the removal of the former, Pfannenstiel's incision was used. Patient received a drain, localized in the left-sided upper abdomen. Operation lasted 88 min, and bleeding was approximately 50 mL. After operation, the patient was taken to the post anesthesia care unit. Patient did well following surgery and was discharged on the 5th postoperative day.

OUTCOME AND FOLLOW-UP

Case 1

Patient was seen again 2 wk after operation. Physical examination showed normal findings, recovery appeared to be proper, and patient felt subjectively healthy. Weight was stable (65 kg, body mass index: 21.2 kg/m^2). Examination of blood sample showed thrombocytosis of 662,000/μL and Howell-Jolly bodies as a sign of an asplenic situation[16,17]. Two weeks later, the patient returned and showed a small wound healing disorder due to a suture granuloma. The wound was re-opened, the suture was removed, and the wound was re-adapted with patches.

Case 2

Patient was seen again 4 wk after operation. Physical examination showed normal findings, recovery appeared to be proper, and patient felt subjectively healthy. Weight was stable (50 kg, body mass index: 22.2 kg/m^2). Measurement of blood samples revealed normal findings with exception of Howell-Jolly bodies as a sign of an asplenic situation[16,17].

DISCUSSION

The occurrence of SANT is very low, with only a few hundred cases world-wide. SANT was first termed in 2004 by Martel *et al*[3], who were able to collect 25 cases of SANT. The histological features of SANT were described years before in a few case reports but never designated as SANT[9]. Although our hospital is a high-volume surgical center, we found only two cases of SANT in our archives between 2004 and 2019. SANT can be well differentiated from other benign lesions histologically[4], but it appears that it is not possible to differentiate SANT clinically. Fine needle aspiration of spleen is necessary due to the risk of peritoneal spreading of a potential malignant lesion and a high risk of post-interventional bleedings is not preferable. Imaging methods often misdiagnose SANT as hemangioma, hamartoma, or, as in one of our cases, malignancy. One case of SANT showed a minimally lower attenuation on unenhanced computed tomography images and hypoattenuation during hepatic arterial and the portal venous phase with an undulating peripheral enhancement

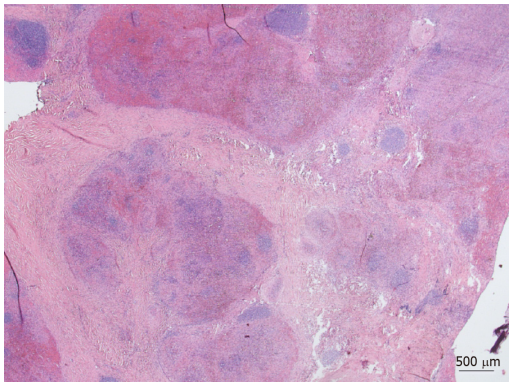

Figure 2 Histological appearance of the first case with typical findings of sclerosing angiomatoid nodular transformation.

compared to normal splenic tissue[6]. Furthermore, contrast enhanced imaging showed nearly an isodense lesion with an unenhanced central stellate area, which is slightly different from other solid masses of the spleen[6]. Hemangioma normally shows a homogeneous and marked contrast enhancement[18]. Differentiation of hamartoma is a bit more difficult as it can be inhomogeneous, showing just a contour abnormality[18]. Further splenic tumors are known, although their radiological diagnoses need not be mentioned in detail here. Generally, the differentiation of solid masses of the spleen is challenging, and misdiagnoses are not surprising due to the variety of splenic tumors.

To our knowledge, nothing is known about the risk of rupture of SANT. Vascular lesions of the spleen, such as hemangioma, are often discussed to be at an increased risk of rupture, and are, therefore, rapidly resected. The recommendation for this procedure is almost 60-years-old[11], and others indicated that it might be possible to observe patients with small, asymptomatic lesions[19]. A systematic review of splenic rupture reports of patients without known risk factors for splenic rupture showed only eight hemangioma and hamartoma cases out of 613 reviewed cases (1.3%) as a reason for rupture[20]. Nevertheless, not only the risk of rupture remains unclear, it is also unclear whether SANT might develop malignant transformation. To date, no case of recurrence of SANT has been reported. Our patients received a short-term after-care of less than 2 years, but they showed no signs of recurrence so far. Surgical resection appears to be a curative response to SANT, restraining the risk of rupture and malignant transformation. Whether it is even necessary, especially for asymptomatic and small cases, remains unclear. As SANT is often an incidental finding, most of the patients are asymptomatic, leading to the question if routine observation might be an alternative. The morbidity rate of splenectomy is approximately 30%, and mortality rate is even up to 15%, depending upon patients' condition and indication for splenectomy[21,22]. Splenectomy has a high risk for intra- and postoperative hemorrhages[22].

The major long-term risk is an overwhelming post-splenectomy infection with an increased risk of fulminant infection, caused by encapsulated bacteria such as *Streptococcus pneumoniae*[17]. Mortality of overwhelming post-splenectomy infection (OPSI) is up to 70%[23]. Nowadays, OPSI is preventable by vaccination, but it is reported that only two-thirds of post-splenectomy patients followed vaccination recommendations and that there are still known deadly cases of vaccination-preventable OPSI[24,25]. Awareness of intra- and postoperative complications of splenectomy is necessary before indicating surgery. There is no doubt that rupture is a life-threatening event, but the risk level of rupture, as well as the risk of malignancy of SANT, remains unclear, and it is, therefore, difficult to estimate the risk-ratio of operation, post-splenectomy living, and occurrence of rupture. It is supposable that most patients were surgically treated due to an unclarified, possibly malignant lesion and not to an occurrence of symptoms. Symptomatic patients might benefit from surgical resection. Difficulty to eat, as stated by our patients, is also reported to be typically observed in patients with SANT[3,5,6]. The symptomatology appears to be plausible in large findings as Figure 2 of Case 2 shows obstructive displacement of the stomach. Severe and acute life-threatening cases of SANT are extremely rare[26], and it is not known if they might be preventable by an early surgical resection. In summary, further research has to focus on improvement of clinical and radiological diagnosis of SANT and on evaluation of the clinical course of SANT. It has to be clarified whether it is possible to observe patients with small and asymptomatic vascular lesions of the spleen or if surgery is always necessary.

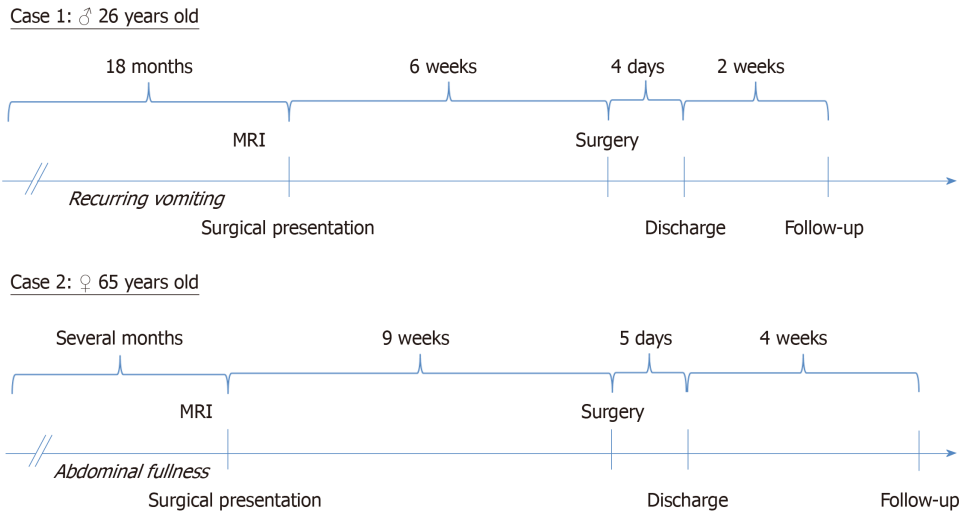

Figure 3 Timeline of both cases.

CONCLUSION

SANT is a rare cause for splenectomy and an incidental histological finding. Some patients develop non-specific gastrointestinal symptoms insecurely related to SANT. Severe cases of SANT are extremely rare. Splenectomy is a promising curative therapeutic approach for SANT. Further research should focus on clinical and radiological diagnosis of SANT as well as on treatment of patients with asymptomatic and small findings.

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