## **Case Report**

# **Preoperative Diagnosis of a Torsioned Accessory Spleen**

Miki Yoshida1\*, Tsukasa Saida<sup>2</sup>, Souta Masuoka<sup>3</sup>, Aiko Urushibara<sup>3</sup>, Fumiko Chiba<sup>4</sup>, Kouji Masumoto<sup>4</sup>

<sup>1</sup>Departments of Radiology, University of Tsukuba Hospital, Tsukuba, Japan, <sup>2</sup>Department of Radiology, Faculty of Medicine, University of Tsukuba, Tsukuba, Japan, <sup>3</sup>Department of Radiology, University of Tsukuba Hospital, Tsukuba, Japan, <sup>4</sup>Department of Pediatric Surgery, Faculty of Medicine, University of Tsukuba, Tsukuba, Japan, <sup>4</sup>Department of Pediatric Surgery, Faculty of Medicine, University of Tsukuba, Tsukuba, Japan, <sup>4</sup>Department of Pediatric Surgery, Faculty of Medicine, University of Tsukuba, Tsukuba, Japan, <sup>4</sup>Department of Pediatric Surgery, Faculty of Medicine, University of Tsukuba, Tsukuba, Japan

# Abstract

We report a case of a 12-year-old boy with an accessory spleen torsion. He presented with left-sided abdominal pain after trauma. A 4 cm oval mass without contrast enhancement was detected on contrast-enhanced computed tomography (CT), and ultrasound (US) showed a 4 cm oval mass below the spleen. The mass mainly consisted of high echoes similar to the spleen; the central part showed irregularly low echoes. Subsequent follow-up daily US examinations showed gradual expansion of the central low echoes with conspicuous hyperechoic dots. Discontinuation of the branch from the splenic artery to the mass was observed, both, on US and CT. These findings led to the diagnosis of a hemorrhagic infarct caused by torsion of the accessory spleen. Laparoscopy showed adherence of the accessory spleen to the omentum and colon by twisting four times around its axis. It was resected and confirmed the diagnosis of a torsioned accessory spleen.

Keywords: Accessory spleen, infarction, torsion, twist, ultrasonography, ultrasound

# INTRODUCTION

The accessory spleen is a congenital anomaly that consists of ectopic splenic tissue separated from the main body of the spleen.<sup>[1]</sup> The frequency of the accessory spleen is about 10%–30% in autopsy series.<sup>[2]</sup>

In general, the accessory spleen is asymptomatic and rarely causes clinical symptoms. However, in some cases, the accessory spleen may present with symptoms related to torsion and infarction. Accessory spleen torsion is a very rare condition often difficult to diagnose. We present a case of accessory spleen torsion that was diagnosed preoperatively with ultrasound (US).

# **CASE REPORT**

An otherwise healthy 12-year-old boy had left-sided abdominal pain after being beaten in the area. He then experienced temporal symptom improvements, but his symptoms recurred, prompting visit to a nearby hospital. The abdominal roentgenogram could not show abnormal findings [Figure 1a]. A mass was detected on contrast-enhanced computed tomography (CT), and he was referred to our hospital forfurther

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investigation and treatment. Physical examination revealed tenderness on the left abdomen. C-reactive protein was 3.6 mg/ dl, and other laboratory tests were normal.

Contrast-enhanced CT showed an oval mass in the upper left abdomen adjacent to the pancreatic tail, clearly separated from the spleen, the left kidney, and the left adrenal gland [Figure 1b]. The mass, located below the spleen, was less dense than the spleen. A tubular low density structure connected to the normal spleen was detected above the mass; thus, it was considered as twisting of the vascular pedicle of the accessory spleen.

US revealed an inhomogeneous oval mass, measuring 4 cm in diameter below the spleen [Figure 2a]. The mass was mainly hyperechoic, similar to the spleen, and the central part showed irregular low echoes. A small amount of ascites was detected in the pelvis. The following day, there was a notable decrease in the internal echo of the mass with linear hyperechoic spots noted in the hypoechoic areas [Figure 2b]. Four days later, the

> Address for correspondence: Dr. Miki Yoshida, Departments of Radiology, University of Tsukuba Hospital, Tsukuba, Ibaraki, Japan. E-mail: myoshida-nii@umin.ac.jp

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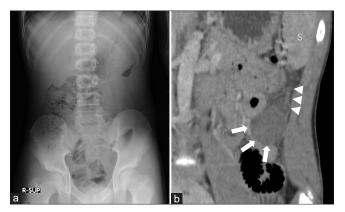
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expansion of the internal hypoechoic areas was noted, and the linear hyperechoic spots became more prominent [Figure 2c]. A tubular structure from the upper edge of the mass to the spleen was detected [Figure 3a and b], and there was no blood flow signal in the mass [Figure 3c]. This tubular structure was considered to be a vascular pedicle of the accessory spleen; thus, accessory spleen torsion was diagnosed.

Twenty-five days after the injury, laparoscopic resection was performed [Figure 4a], and the accessory spleen was found adhered to the omentum and colon, twisted four times around its axis. The cut surface of the excised specimen showed a red splenic tissue [Figure 4b], and pathological examination revealed hemorrhagic infarction of the accessory spleen, confirming the diagnosis of accessory spleen torsion. His postoperative course was uneventful and no abnormal findings in laboratory data. The patient was discharged on the second postoperative day.

### DISCUSSION

Seventy-five percent of accessory spleens are located in the splenic hilum, 20% near the tail of the pancreas, and the remaining 5% are found in the splenic ligaments, the omentum, scrotum, and rarely, the mediastinum.<sup>[1,3]</sup> The size of accessory spleens varies from a few millimeters up to 1.5



**Figure 1:** (a) The abdominal roentgenogram shows no abnormality. (b) Contrast-enhanced computed tomography shows a hypodense 4 cm mass (arrows). There is a non-enhancing tubular structure extending from the upper edge of the mass to the spleen (arrowheads). The spleen is located in the normal position (s)



**Figure 3:** US findings four days later (Toshiba Aplio 500) (a and b) Ultrasound images on left intercostal scan. A tubular structure (arrows) without blood flow signal expanding from the upper edge of the mass to the spleen is detected (Advanced Dynamic Flow). The tubular structure disappears on the spleen side. (c) Blood flow signal is not detected in the mass even on Superb Microvascular Imaging

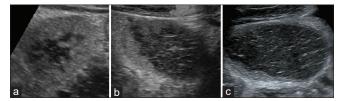
to 2 cm in diameter.<sup>[1,2]</sup> The vascularization of the ectopic tissue is supplied by the arterial branches arising from the splenic artery.<sup>[4]</sup>

In general, the accessory spleen is asymptomatic and rarely causes clinical symptoms. Torsion and infarction of an accessory spleen is a very rare occurrence. Affected patients range from infants to the elderly, with more than half of the reported cases in children.<sup>[5]</sup> The symptoms vary from vague abdominal pain seen in wandering accessory spleen with intermittent torsion, to fever, vomiting, and acute onset of severe abdominal pain, observed when there is an infarction.<sup>[5,6]</sup>

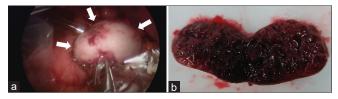
Preoperative diagnosis of accessory splenic torsion is difficult, and in most previous cases, the diagnoses were not made preoperatively. In some cases where a preoperative diagnosis was possible, the diagnoses were established with the aid of imaging.

The US descriptions of previously reported accessory splenic torsion include a hypoechoic well-encapsulated oval mass in the left upper quadrant,<sup>[5]</sup> and a nodular solid mass below the splenic hilum, adjacent to the left kidney.<sup>[6]</sup> Doppler imaging could be used to evaluate the degree of vascularization of the mass.<sup>[5]</sup> The contrast-enhanced US is also useful.

Torsion causes infarction and splenic infarction also occurs in patients with myeloproliferative syndromes, hemolytic anemias, hematological malignancy, vasculitis, cardiac emboli, portal hypertension, and infiltrative disorders. According to previous reports, splenic infarctions appear as subcapsular and triangular hypoechoic segmental lesions, with the bases parallel to the splenic periphery and the apices pointing toward the hilum on the US. On color doppler US, the affected areas show



**Figure 2:** Ultrasound findings with time (a) (Hitachi Aloka Prosound  $\alpha$ 7) ultrasound examination on the first day shows an isoechoic mass when compared with the normal spleen in the left abdomen. The mass has an irregular central hypoechoic area. (b) (Hitachi Aloka Prosound  $\alpha$ 7) The next day, expansion of the central hypoechoic area is noted and emerging linear hyperechoic spots are noted. (c) (Toshiba Aplio 500) Four days later, further expansion of the hypoechoic area with internal linear hyperechoic spots are noted



**Figure 4:** (a) The twisted accessory spleen is laparoscopically dipicted as a well-defined mass (arrows) (b) The resected specimen shows a hemorrhagic infarction

no blood flow. In addition, US findings of splenic infarction change from isoechoic with normal spleen immediately after onset, to low echo after 24 hours, and inhomogeneity months later as scar states.<sup>[7,8]</sup> These findings are known to be seen in accessory spleen torsion.

It has been reported that there is a high possibility of relapse in spontaneous remission cases. Therefore, in patients with symptoms, resection of the accessory spleen is considered as soon as possible. Laparoscopy has been tailored to resect smaller sized accessory spleens, diagnosed preoperatively, and has been gradually modified to resect larger accessory spleens.<sup>[9]</sup>

In our case, the accessory spleen was 4.5 cm, larger than usual, and considered a risk of torsion. The initial US image showed marginal hyperechoic areas of the mass with internal hypoechoic areas without blood flow. These findings suggested >24 hours postinfarction due to torsion. In addition, we could observe a tubular structure considered as a twisted vascular pedicle of the accessory spleen, thereby confirming the diagnosis.

To the best of our knowledge, this is the first report in which changes to the internal echo of a torsioned accessory spleen could be observed with time. Observing these changes over time is useful for the diagnosis.

Accessory spleen torsion is a very rare condition, and it is often difficult to diagnose. In this case, using US and CT, we could detect a tubular structure extending from the upper edge of the mass to the spleen, suggesting accessory spleen torsion. Subsequent US exams showed internal echo changes of the mass with time, confirming the diagnosis of infarction of the accessory spleen due to torsion.

#### **Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's guardian has given the patient's consent for his images and other clinical information to be reported in the journal. The patient's guardian understands that the patient's name and initial will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

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Nil.

#### **Conflicts of interest**

There are no conflicts of interest.

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