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CASE REPORT

Mesenteric lipoblastoma presenting as a small intestinal volvulus in an infant: A case report and literature review



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Yuka Nagano, Keiichi Uchida*, Mikihiro Inoue, Shozo Ide, Tadanobu Shimura, Kiyoshi Hashimoto, Yuki Koike, Masato Kusunoki

Department of Gastrointestinal and Pediatric Surgery, Mie University Graduate School of Medicine, Tsu, Mie, Japan

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KEYWORDS lipoblastoma; mesenterium; volvulus **Summary** A 1-year-old boy with no underlying disorder presented with non-bilious vomiting since 4 days before admission. He was referred to our hospital and was diagnosed with a small bowel obstruction due to an intraabdominal tumor. Laparotomy revealed an intestinal volvulus with a soft and lobulated tumor arising from the mesentery. The resected tumor with a small part of the small bowel was diagnosed as lipoblastoma histologically. From a literature review, mesenteric lipoblastoma with an intestinal volvulus showed different characteristics such as greater frequency of vomiting and less frequency of abdominal mass as clinical symptoms, and the size of the tumor was smaller than that of the tumor without the intestinal volvulus. Copyright © 2013, Asian Surgical Association. Published by Elsevier Taiwan LLC. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

1. Introduction

Lipoblastoma is a benign soft tissue tumor originating from embryonal white fat tissue. Most cases are found in infants and young children, especially in the age of 3 years or younger.^{1,2} The tumor frequently occurs in the subcutaneous tissues of the extremities and the trunk.^{3–5} It can also arise in the retroperitoneum, abdomen, chest, mediastinum, scrotum, and so on.^{6–8} Among these, mesenteric occurrence is rare, and only 19 cases have been reported to date. In this report, we present an additional case of mesenteric lipoblastoma developing into a small intestinal volvulus and also review the reported cases with mesenteric lipoblastoma.

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^{*} Corresponding author. Department of Gastrointestinal and Pediatric Surgery, Mie University Graduate School of Medicine, Edobashi 2-174, Tsu, Mie 514-8507, Japan.

E-mail address: ucchie@clin.medic.mie-u.ac.jp (K. Uchida).

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2. Case report

A 1-year-old boy with no underlying disorder presented with nonbilious vomiting since 4 days before admission. He visited a hospital because the vomiting has been getting bilious and more frequent with abdominal distension. Imaging tests showed small intestinal obstruction due to the mass lesion, and he was referred to our hospital to further intervention. On admission, he looked pale and lifeless. On palpation, the abdomen was markedly distended without peritoneal signs and an elastic, soft, and mobile mass was detected in the right lower abdomen with a measurement of 10 cm \times 5 cm. Laboratory investigations revealed mild anemia, hyponatremia, and hypochloremia without other remarks. Abdominal X-ray film showed small bowel obstruction (SBO) (Fig. 1A). Abdominal computerized tomography (CT) scan revealed dilated small bowel and a heterogeneous mass without calcification (Fig. 1B). Patient underwent an emergent laparotomy with a preoperative diagnosis of SBO due to an intraabdominal tumor containing fat tissues. On operation, it was noticed that a soft and lobulated tumor with a smooth, glistening capsule arose from the mesentery of ileum, and small intestine was rotated 360° around the tumor (Fig. 1C). An additional 360° rotated small intestinal volvulus around the proximal mesentery was also found. Although there was no sign of necrosis in the small intestine, the tumor severely adhered to the border of the small intestines, and it was difficult to remove the tumor without bowel resection. We resected the tumor *en bloc*, including the small bowel, which was 15 cm in length. Histological findings were compatible with those of lipoblastoma without evidence of tumor invasion to the bordering intestine. The postoperative course was uneventful, and he was discharged from the hospital on postoperative day 10. There is no evidence of recurrence on clinical symptom and magnetic resonance imaging (MRI) examination for 1 year after operation.

3. Discussion

The lipoblastoma is rare, benign tumor composed of fetal adipose tissue.⁹ Most of them are seen in infants and young children, and 80-90% of cases occur at 3 years of age or younger.^{1,2} Boys are affected two to three times more often than girls.¹⁰ Lipoblastoma is commonly located in the extremities^{3–5} and also in the body trunk, head and neck, mediastinum, inguinal region, scrotum, buttock, perirectal area, retroperitoneum, and rarely the mesentery.^{6–8}

Abdominal lipoblastomas represent roughly 7% of all lipoblastomas, and most lipoblastomas arise in the retroperitoneum.^{3,4,11} Among 24 studies that reported the cases arising in the mesentery, 18 cases were described in detail.^{9,12–26} Then, we reviewed 19 cases, including our additional case in this study, and investigated whether the clinical difference in mesenteric lipoblastoma complicated the intestinal volvulus or not (Table 1). Statistical analysis

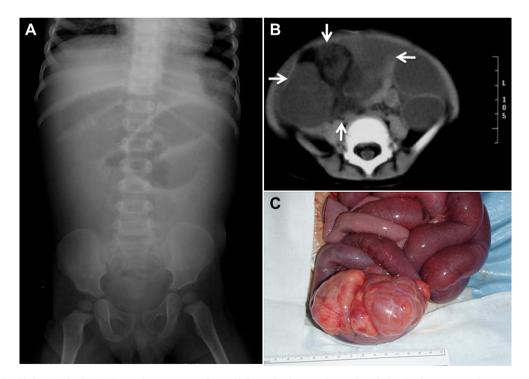


Figure 1 (A) Abdominal plain X-ray demonstrated small bowel obstruction. (B) Abdominal computed tomography revealed dilated small bowel and a heterogeneous mass without calcification. (C) Intraoperative photograph shows a soft and lobulated tumor with smooth, glistening capsule arising from the mesentery 180 cm distal to the Treitz' ligament. It shows that the small intestine has rotated 360° around the tumor, and an additional 360° rotated small intestinal volvulus around the more proximal mesentery was also found.

	Volvulus group $(n = 3)$	Nonvolvulus group ($n = 14$)	p value
Gender (boy/girl)	2/1	9/5	n.s.
Age (mean \pm SD, year)	$\textbf{3.3}\pm\textbf{3.2}$	2.0 ± 1.2	n.s.
Symptoms at presentation to hospital			
Abdominal mass or distension	0 (0%)	10 (71%)	p = 0.025
Abdominal pain	1 (33%)	4 (29%)	n.s.
Vomiting	3 (100%)	2 (14%)	p = 0.0031
Location			
Jejunum	2 (66%)	2 (14%)	n.s.
Ileum	1 (33%)	5 (36%)	
Transverse colon	0 (0%)	1 (7%)	
Unknown	0 (0%)	6 (43%)	
Maximum diameter of the tumor	$9.7 \pm 1.5, 8 - 11$	15.3 \pm 3.6, 9.7–23	p = 0.024
(mean \pm SD, range, cm)			
Intestinal resection (yes/no)	2/1	6/8	n.s.

Table 1 Comparison of mesenteric lipoblastoma with or without volvulus in the literature

was performed using Statview 5.0 for Windows (SAS Institute Inc., Cary, NC). Values for continuous variables were expressed as mean \pm 1 standard deviation. The Mann-Whitney *U* test and Fisher's exact Chi-square test were used to compare the observed differences between the groups; *p* < 0.05 was considered statistically significant.

A small intestinal volvulus was found in three cases, including our present case^{9,14}; however, there is no case associated with intestinal necrosis. There was no difference in gender and age at onset between the volvulus and nonvolvulus groups. Considering the patients' symptoms on visiting the hospital, patients in the volvulus group revealed an abdominal mass less frequently and vomiting more frequently than those in the nonvolvulus group, which were statistically significant (p = 0.035 and 0.0055, respectively). There was no difference in the location of the tumor between both groups. The diameter of the lipoblastoma in the volvulus group was significantly smaller than that of the lipoblastoma in the nonvolvulus group (8.8 \pm 1.0 cm, 8–10 in range vs. 15.4 \pm 3.3 cm, 11–23 in range, p = 0.0094). Although the lipoblastoma is a benign tumor and only complete tumor resection is the effective procedure, 9 out of 19 (47%) cases needed additional bowel resection because of severe tumor adhesion to the border of the intestine such as in the present case. There is no difference in intestinal resection rate between the volvulus and nonvolvulus groups. No recurrence cases have been reported in the mesenteric lipoblastoma to date in the literature.

Lipoblastoma arising from the mesentery is extremely rare, and cure is expected by complete resection of a tumor with or without a part of the intestine. Mesenteric lipoblastoma with an intestinal volvulus had different characteristics such as more frequency of vomiting and less frequency of abdominal mass as clinical symptoms and the size of the tumor was smaller than that of the tumor without the intestinal volvulus.

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