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A case of cystic hemangioma in mesentery

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ABSTRACT

A nine-year-old, otherwise healthy girl presented with abdominal pain. On exam, there was a palpable cystic tumor in the left abdomen. According to imaging examinations, we tentatively diagnosed the tumor as lymphangioma of mesentery with bleeding. We planned to perform laparoscopic surgery to remove the tumor. On laparoscopy, cyst aspiration was performed to reduce the volume of the cysts, and the aspirated fluid was found to contain red blood cells. Due to technical difficulties, laparoscopic surgery was converted to open surgery. RBCs were detected in the cyst fluid at the sigmoid colon and intestinal mesentery. Resection of the cystic tumor and sigmoidectomy were performed. On pathologic examination of the tumor, there were multiple cysts but there was no cavernous part. The largest cyst was 6 × 5 × 3 cm. Simple squamous epithelium cells were found along parts of the inner lining of enlarged luminal cysts. On immunohistochemical staining, the tumor was positive for CD31 and CD34, and negative on D2-40 antibody staining. Therefore, we diagnosed this specimen as hemangioma with giant cysts at the mesentery. We conclude that bleeding into cysts of a hemangioma of mesentery might lead to the development of giant cysts and, in turn, a cystic tumor in the abdomen.

1. Introduction

Cystic and cyst-like abdominal masses in children can be difficult to characterize. Mesenteric cysts are generally cystic lymphangiomas found in the small bowel and large bowel mesentery [1]. We experienced a case of cystic hemangioma with bleeding in mesentery.

2. Case report

2.1. Patient history

The patient was an otherwise healthy, nine-year-old girl with constipation from 8 years of age. She had abdominal pain without a bowel movement for 5 days, and glycerin enema was performed by her mother at home. The unprecedented continuous abdominal pain gradually worsened after glycerin enema day by day, and the patient was brought to our outpatient clinic. On physical examination, the patient's abdomen was slightly distended. An abdominal tumor was palpable at the left middle part of the abdomen. Abdominal X-ray confirmed the presence of a tumor, although dilation of the ileum and colon due to gas was not seen (Fig. 1). Ultrasonography revealed an echogenic tumor of 12 cm in size with a septum (Fig. 2). The tumor had multiple cysts. Color doppler ultrasonography did not detect blood flow in the tumor wall. Computed tomography (CT) showed a cystic tumor with multiple cysts and a small high-density area, and there was no enhancement at the tumor site. The density in the tumor was slightly higher than the density of water (Fig. 3). Magnetic resonance imaging (MRI) detected a lobulated tumor of 11.5× 7.2 × 5.6 cm in size with a septum in the tumor. Both T1-and T2-weighted MRI revealed high intensity signals (Fig. 4). We tentatively diagnosed this tumor as cystic lymphangioma with bleeding. The family and patient chose tumor resection by laparoscopic surgery due to continuous abdominal pain.

2.2. Operative findings

We planned to remove the abdominal tumor by laparoscopic surgery. We confirmed the presence of a dark-red cystic mass by laparoscopy (Fig. 5). First, cyst aspiration was performed to reduce

Abbreviations: MRI, magnetic resonance imaging; CT, computed tomography.

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Fig. 1. Abdominal X-ray. Abdominal X-ray confirmed the presence of a tumor, although dilation of the ileum and colon due to gas was not seen.

the volume of the cysts. Ninety ml of serous fluid that was found to contain red blood cells (4.4 million/μL) was aspirated from the cystic tumor. The cystic tumor was located at the sigmoid colon mesentery and intestinal mesentery. Due to technical difficulties, the laparoscopic operation was converted to open operation and the wound incision was elongated. We observed that there was a large defect in the sigmoid colon mesentery, and the cystic tumor and part of the sigmoid colon were excised.

2.3. Pathological findings (Fig. 6)

The tumor consisted of multiple cysts composed of fibrous tissue. The maximum size of a cyst was 6 × 5 × 3 cm. The presence of simple squamous epithelium cells was confirmed at parts of the inner lining of the enlarged luminal cysts. There were multiple cystic components and no cavernous part in the dissected specimen. On immunohistochemistry, these simple squamous epithelium cells were positive for CD31 and CD34, which are specific markers of hemangioma, but were not stained with D2-40 antibody which stains for a specific marker of lymphangioma.

Therefore, we confirmed that these cells were endothelial cells, and diagnosed this specimen as hemangioma with giant cysts at mesentery.

3. Discussion

The incidence of mesenteric cystic masses has been estimated at 1/100,000 in the United States [2]. Most mesenteric masses are cystic lymphangioma [2]. In general, cystic lymphangioma in the abdomen should be differentiated from enteric cyst, intestinal duplication, mesothelioma, dermoid cyst, and mucinous cyst [3]. The typical cystic lymphangioma is detected as a low echoic area on ultrasonography. It is visualized as a low-to middle-intensity area on T1-weighted MRI, and as a high-intensity area on T2-weighted MRI. The tumor in our case was visualized as an echogenic tumor without rich blood flow by color doppler and/or ultrasonography, and high-intensity imaging at the tumor was shown by T1-and T2-weighted MRI. Therefore, we initially diagnosed this cyst as lymphangioma with bleeding. A hemangioma tumor is visualized as a low echoic mass and has rich blood flow on color doppler and/or ultrasonography and as a high-intensity mass on T1-and T2-weighted MRI.

Hemangiomas in the gastrointestinal system of children are of capillary, cavernous or mixed type in histopathology [4]. Eight cases of hemangioma involving the mesentery were presented as “big or large neoplasms” in the English literature to our knowledge [5–12]. Five of the cases involved a large cystic lesion [5–8,12]. However, the definition of what constitutes a “large cyst” or “giant cyst” in the mesentery had not been given. According to previous reports of large cyst hemangioma involving a central neuron, the maximum diameter of the cystic lesions was more than 20 mm [13,14]. Only one of the reported cases of cavernous and venous mixed type hemangioma in the mesentery was in a child, who was 5 years of age [12]. Our case is only the second case report of a cystic hemangioma in mesentery in a child. It has been speculated that cystic hemangioma occurs by recurrent hemorrhage from a vascular malformation or nanocapillary and the osmotic transport of fluid into the cyst [13,14]. We speculate that bowel movements caused by glycerin enema led to bleeding into cysts of the hemangioma, which led to the development of giant cysts and eventually a tumor and abdominal pain in our case. Five previously-reported patients with hemangioma in mesentery with cystic formation had abdominal pain [5–8,12], some of whom had progressively worsening pain.

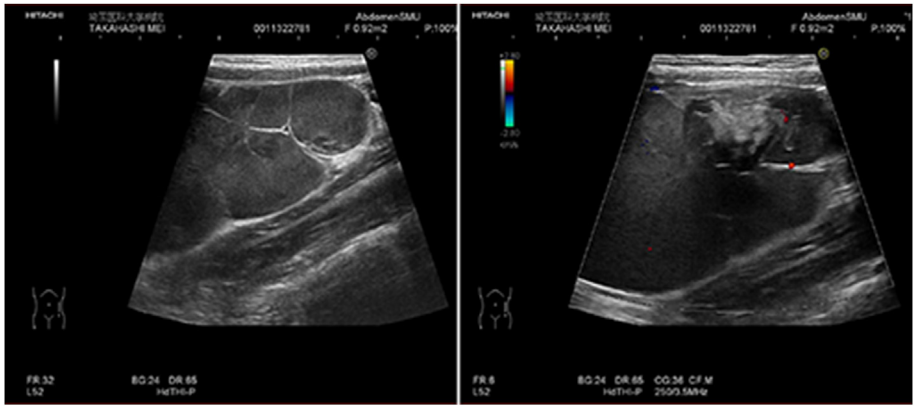

Fig. 2. Abdominal ultrasonography. Ultrasonography revealed an echogenic tumor of 12 cm in size with a septum (Fig. 2). The tumor had multiple cysts. Color doppler ultrasonography did not detect blood flow in the tumor wall.