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An unusual cause of paediatric abdominal pain: Mesenteric masses accompanied with volvulus

BOWEL

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Original Article

ABSTRACT

Background/Aims: Volvulus caused by mesenteric masses is rare and may result in serious consequences. This study aimed to better characterize volvulus caused by mesenteric masses in children.

Materials and Methods: A retrospective study was conducted in 24 patients who underwent surgical treatment between January 1994 and January 2014 in one single institution.

Results: There were 10 boys and 14 girls. The most frequent findings were abdominal pain (100%), emesis (91.7%) and nausea (83.3%). Physical examination showed positive ileus signs in majority cases, and palpable mass was found in half of the patients. Ultrasound and CT scans revealed mesenteric masses in 21 and 24 patients, and 'whirlpool sign' was observed in 19 and 22 patients, respectively. Emergency laparotomy was performed in all patients. Histological examination revealed that 18 cystic masses were lymphangioma, 5 solid cases were lipoma and the remaining one was lipoblastoma. The postoperative course was uneventful in 22 patients, and postoperative obstruction and incision infection occurred in 2 patients. There was no evidence of recurrence at follow-up.

Conclusion: Volvulus caused by mesenteric masses is a rare but potentially life-threatening cause of abdominal pain, which should be considered in the differential diagnosis of paediatric acute abdominal pain.

Keywords: Abdominal pain, mesenteric masses, volvulus, paediatric

INTRODUCTION

Acute abdominal pain is one of the most common complaints in children, and it is a diagnostic challenge for doctors because of the variety of underlying causes. Majority of acute abdominal pain is due to intestinal spasms, which are usually a self-limiting and benign condition, such as during constipation, viral illness or gastroenteritis (1). However, identifying children who have uncommon and potentially life-threatening conditions that require urgent treatment is a big challenge, such as volvulus, intussusception, appendicitis or intestinal adhesion (1,2). Intestinal volvulus is a rare but life-threatening surgical emergency; nevertheless, intestinal adhesions, ascariasis and mesenteric mass (cyst or solid) are potential predisposing factors of volvulus. However, mesenteric masses, either cyst or solid, are quite rare in paediatrics. In addition, the presentations maybe variable, ranging from asymptomatic to intermittent abdominal pain, vomiting or peritonitis. Their symptoms and physical findings vary and are frequently nonspecific. Large lesions can cause obstructive symptoms due either to mass compression or, rarely, to volvulus (3,4). Therefore, preoperative diagnosis is often difficult. The definitive diagnosis is frequently made at the time of abdominal exploration or postmortem examination (5). Therefore, it is very important for early diagnosis to avoid bowel necrosis and intestinal perforation. To better characterize the clinical features of mesenteric masses accompanied with volvulus, we reviewed all cases treated surgically in our institution over the last 20 years.

MATERIALS AND METHODS

With ethics approval from the hospital's IRB, we conducted a retrospective study to determine the clinical characteristics and laboratory findings of children (age

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<18 years) with volvulus caused by mesenteric masses at The Children's Hospital of Chongqing Medical University hospital between January 1994 and January 2014. Emergency colour Doppler ultrasound (CDUS) and abdominal plain film and computed tomography (CT) scans were performed for every patient and interpreted by experienced paediatric radiologists. The case notes were examined in detail, including the patient's age; duration of symptoms before consulting; main symptoms; associated symptoms; inducing factors and the physical examination findings. Laboratory and radiographic data included complete blood count (CBC), serum electrolytes, CDUS and enhanced CT scan.

Emergent laparotomy was performed for all patients, and resection of the affected segment of the intestine followed by end-to-end anastomosis, marsupialization with intima cauterization and enucleation of the tumour was performed according to the location of the mass and the vitality of the intestine after derotation. Follow-up was determined in the out-patient department by post-discharge appointments with a physician at our institution.

The statistical analyses were performed using the Statistical Package for the Social Sciences software version 23.0 (SPSS Inc.; Chicago, IL, USA). Categorical data between groups were compared using the chi-square test. Results were considered statistically significant if analysis yielded a p value <0.05.

RESULTS

Patients

A total of 24 children (median age 5 years; range 0.5–14 years; 14 girls and 10 boys) with mesenteric masses accompanied with volvulus were surgically treated at our institution between 1994 and 2014. None had family history. Demographics and diagnosis are demonstrated in Table 1.

Clinical signs

The overall incidences of clinical findings and the results of laboratory and radiological studies are listed in Table 2. Only two patients had a history of trauma or activities. Most patients sought for medical advice within 1 day, which accounted for 63.3% of all children. However, delayed admission to the hospital was also considerable; four patients came to the hospital more than 1 week since he/she had developed symptoms, and these patients were all 'left-behind children' (which meant their parents have gone out to work in the city and they were raised by grandparents) and came from rural areas. The most severe case was a 14-year-old girl who had a history of recurrent abdominal pain for 2 years accompanied by chronic constipation for a long time. The most frequent finding was abdominal pain, emesis and nausea, which was presented in 24 (100%), 22 (91.7%) and 20 (83.3%), respectively. In addition, constipation, fever and bleeding per rectum were also observed in our series. Physical examination showed positive ileus signs (abdominal distention, abdominal tenderness, diminished bowel sounds) in majority cases, and palpable mass was only found in half of the patients.

Table 1. Demographics and diagnosis of 24 children

	N	% of total
Gender		
Male	10	41.7
Female	14	58.3
Age (Years)		
0–3	10	41.7
4–6	8	33.3
7–10	4	16.7
11–18	2	8.3
Diagnosis		
Lymphangioma	18	75
Lipoma	5	20.8
Lipoblastoma	1	4.2
Region		
Urban area	10	41.7
Rural area	14	58.3

N: number

Table 2. Clinical Findings of mesenteric masses accompanied with volvulus

	Number and Incidence		Number and Incidence
History of trauma or activities	2 (8.3%)	Nausea	20 (83.3%)
Duration of symptoms (d)		Constipation	8 (33.3%)
<1 d	16 (66.7%)	Fever	3 (12.5%)
1–7 d	4 (16.7%)	Bleeding per rectum	1 (4.2%)
7–14 d	2 (8.3%)	Palpable mass	12 (50%)
>14 d	2 (8.3%)	Abdominal distention	16 (66.7%)
Abdominal pain	24 (100%)	Abdominal tenderness	18 (75%)
Emesis	22 (91.7%)	Diminished bowel sounds	13 (54.2%)

Table 3. Results of laboratory and radiologic studies of mesenteric masses accompanied with volvulus

	Number and Incidence		Number and Incidence
WBC >10^9/L	13 (54.2%)	Abdominal plain film	
Electrolyte disorder	19 (79.2%)	Bowel obstruction	20 (83.3%)
Abnormal albumin	2 (8.3%)	Dynamic change	4 (16.7%)
CDUS		Abdominal CT	
Cystic multiloculated mass	17 (70.8%)	Intraperitoneal cyst	18 (75%)
Homogenous echogenic mass	4 (16.7%)	Non-enhanced, fat-density mass	6 (25%)
Inconclusive owing to intestinal distension	2 (8.3%)	Whirlpool sign	22 (91.7%)
Whirlpool sign	19 (79.2%)	Abnormal position of SMA and SMV	2 (8.3%)

WBC: white blood cell; CDUS: colour Doppler ultrasound; SMA: superior mesenteric artery; SMV: superior mesenteric vein; CT: computed tomography

Laboratory and radiological studies

The biochemical and radiological data are listed in Table 3. Several indexes were notable. Electrolyte disorder was confirmed in 19 patients, including hyponatremia, hypokalaemia and hypochloraemia. More than half of the patients had elevated white blood cell count. Hypoalbuminemia was found in two patients, both of whom had a long history of chronic abdominal pain and emesis.

Colour Doppler ultrasound revealed cystic multiloculated mass, and homogenous echogenic mass was observed in 17 out of 18 lymphangioma patients (94.4%) and in 4 out of 6 lipoma/lipoblastoma patients (66.6%). The ultrasound results of two patients (1 lymphangioma and 1 lipoma) were inconclusive owing to intestinal distension, and the last patient was misdiagnosed as mesenteric fat. 'Whirlpool sign' was observed in 19 patients; 16 out of 19 cases were lymphangioma and the rest were lipoma.

X-ray examination including abdominal plain film and enhanced CT

Bowel obstruction was found in 20 patients when abdominal plain film was performed. Abdominal enhanced CT showed thin-wall cystic mass, with cystic wall and internal division intensified after contrast in all 18 lymphangioma patients and non-enhanced, fat-density mass in 6 lipoma/lipoblastoma patients. Positive 'Whirlpool sign' was observed in 22 patients, and the other two patients manifested as slightly abnormal position of superior mesenteric artery (SMA) and superior mesenteric vein (SMV).

Operative findings and pathology

Emergency laparotomy was performed after laboratory and radiological examinations in all patients. No bowel necrosis was found in the 16 patients who came to hospital within 1 day; 3 out of the other 8 patients who had a delayed admission to the hospital were confirmed to have bowel necrosis ($p<0.05$). It was noticed that 14 tumours originated from the mesentery of the ileum (12 cystic and 2 solid), 6 tumours were located at the jejunal mesentery (4 cystic and 2 solid), 2 masses were located at the ileocecal junction (1 cystic and 1 solid) and the rest originated from the sigmoid mesocolon (1 cystic and 1 solid). Resection of the affected segment of the intestine followed by end-to-end anastomosis was performed in 18 patients (15 cystic and 3 solid), simple tumour resection (enucleation of tumour) was performed in 4 patients (3 solid and 1 cystic) and marsupialization with intima cauterization was performed in 2 patients (both were cystic). Histological examination revealed 18 cystic masses were lymphangioma, 5 solid cases were lipoma and the remaining one was lipoblastoma.

Postoperative care and complications

Fasting and total parenteral nutrition were administrated in all patients who underwent enterectomy and end-to-end anastomosis for at least 5 days. Enteral nutrition was administered when the intestinal function recovered. The postoperative course was uneventful in 22 patients, and they were discharged from the hospital on postoperative day 7–10. One patient was complicated by an episode of postoperative obstruction, which settled with conservative treatment for 5 days. The last one was complicated by incision infection on the third day after surgery, and antibiotics were given for 1 week. No complications of postoperative haemorrhage, anastomotic fistula and intra-abdominal infection were observed.

Follow-up

Long-term follow-up was achieved in all patients. Follow-up time ranged from 1 to 16 years, with a mean time of 67 months (5.6 years). Radiological examination was performed in all patients at least once every year. There is no evidence of recurrence on clinical symptoms and radiological examinations at follow-up.

DISCUSSION

Intestinal volvulus is a relatively uncommon but potentially life-threatening cause of abdominal pain. The etiology may be primary or secondary. Primary volvulus occurs in a normal abdominal cavity and is very rare. Secondary volvulus often has a predisposing factor, such as adhesions, ascariasis and mesenteric cysts or tumours (6).

Mesenteric masses are rare and consist of a heterogeneous group of lesions (7,8). They can be classified as solid or cystic, benign or malignant; the benign ones are more common than malignant tumours (9). Mesenteric masses are usually discovered incidentally or during investigation of non-specific symptoms (10); these non-specific symptoms are variable, including abdominal pain, nausea and vomiting, constipation and diarrhoea. Symptomatic mesenteric masses are associated with tumour size, tumour location and complications, including infection, rupture, intestinal obstruction, intra-tumour or intra-abdominal haemorrhage and rarely volvulus.

The most common cystic mesenteric masses is lymphangioma (11). It appears to result from congenital malformation of lymphatic vessels rather than a true lymphatic tumour (12,13) and accounts for half of mesenteric cystic masses, with an incidence of 1/200,000 in the United States (10,14). In the mesentery, they can occur at the root or encroach on the lumen and may cause intestinal loops pressing, but due to their nature and liquid nature of small intestinal contents, which usually can pass through, obstructive symptoms may not be presented (15). Most patients may present with abdominal pain, intermittent vomiting or increased abdominal girth, especially within the first 2 years of life. Other uncommon symptoms include weight loss, anorexia, sensation of fullness and constipation. Haemorrhage may cause acute severe pain with rapid expansion of the mass. Rare cases of intestinal obstruction, volvulus and acute pancreatitis from larger lesions have also been described (3,4).

Volvulus caused by mesenteric masses is more rare in paediatric populations, which may result in a closed loop obstruction and may cause complications such as venous and arterial occlusion, resulting in ischemia and necrosis of the bowel, which will result in perforation of the bowel, leading to peritonitis. In our study, abdominal pain, emesis and nausea were the most frequent symptoms, which presented in 24 (100%), 22 (91.7%) and 20 (83.3%) patients, respectively. Palpable mass was only found in half of the patients. However, the clinical presentation is also non-specific, and no clinical sign consistently identifies volvulus. These symptoms may be confused with other acute abdominal diseases, and some patient's fever and mucous bloody diarrhoea might have resulted from secondary bowel inflammation related to volvulus or from unrelated infectious diarrhoea. It has been reported that severe abdominal pain inconsistent with the physical findings is the classic feature of intestinal ischemia (16); however, the degree of abdominal pain was not easy to confirm and the physical examination was inaccurate due to weak compliance in early age children; therefore, accessory examination was essential to rule out other situations.

There are no specific laboratory findings for these patients, but it is important for us to understand the homeostasis. Electrolyte disorder, including hyponatremia, hypokalaemia and hypochloraemia, may be observed due to recurrent vomiting/nausea and anorexia. Anaemia is comparatively uncommon due to the short history, unless intratumoural haemorrhage occurred. Hypoalbuminemia may also exist for long-time vomiting and inappetence. Radiological features on plain film are often non-specific, which show bowel obstruction with distended loops. Ultrasound can detect mesenteric masses as a homogenous echogenic mass or cystic multiloculated masses; however, it may be misdiagnosed as mesenteric fat because the mesentery is rich in fat or inconclusive owing to intestinal distension. As in our series, the ultrasound results of 2 patients (1 lymphangioma and 1 lipoma) were inconclusive owing to intestinal distension, and 1 patient was misdiagnosed as mesenteric fat. The classic sonographic and colour Doppler sign of small-bowel volvulus is the whirlpool sign (17), which corresponds to a clock-wise wrapping of the SMV and the mesentery around the SMA. The whirlpool sign directly indicates the anatomic alteration caused by midvolvulus. The sensitivity of this sonographic sign ranges from 89% to 100% (18,19). CT scans are considered to be more useful in making an accurate diagnosis, especially with CT depicting loops of small bowel forming a whirl-like pattern, and the sensitivity was higher than ultrasound. In a recent prospective trial, the overall sensitivity and negative predictive value of CT scan in the diagnosis of strangulation and ischemia were 96% and 99%, respectively (20).

The recommended treatment of mesenteric masses accompanied with volvulus was emergent laparotomy. After derotation, the procedures depend on the nature of the tumour, the location of the mass and the vitality of loops. At surgery, the operative intention is to remove the entire tumour conservatively, while avoiding damage to the mesenteric vasculature. Simple enucleation is sufficient for well-circumscribed tumours. In some situations, the tumour severely adhered to the border of the small intestines, and it was difficult to remove the tumour without bowel resection. In this situation, resection of the affected segment of the intestine followed by end-to-end anastomosis may be the only choice (21). In some cases, mostly seen in lymphangioma, complete excision is not feasible due to the tumour located near the vital structures or extensive disease. In these cases, drainage of tumour and cauterization of lining using iodine tincture could be an alternative option in those instances.

The incidence of bowel necrosis may depend on the duration of pain at presentation and the degree of the gut tortured. In our series, no bowel necrosis was found in 16 patients who came to the hospital within 1 day. However, 3 out of the other 8 patients who came to the hospital over 1 day were confirmed to have bowel necrosis; thus, early diagnosis and timely treatment is of great importance. Interestingly, there was no evidence of bowel necrosis of the patient who had the longest history. We speculated that volvulus may be incomplete and spontaneous detorsion occurred with relief of obstructive symptoms. For these patients with long-time abdominal pain, careful physical examination and relevant accessory examination should be considered.

Some limits also existed in our study. This is a retrospective study of one single-institution, and due to the low economic status in southwest China, most children coming from rural areas are 'left-behind children', who are raised by their grandparents. Delayed admission to hospitals is very common for this population; thus, some conclusion may not reflect the real situation in China. Multi-centre studies are needed to better understand this special acute abdomen in paediatric populations.

In conclusion, volvulus caused by mesenteric masses is a rare intra-abdominal lesion, which may pose diagnostic difficulties due to its vague non-specific symptoms and its rarity. Therefore, physicians should be aware of the special manifestations of mesenteric masses and avoid inappropriate managements. Our report should serve to remind us to include mesenteric masses accompanied with volvulus as a differential diagnosis of paediatric acute abdominal pain.

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