Case Report

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Website: www.afrjpaedsurg.org
DOI: 10.4103/0189-6725.115050
Quick Response Code:

Omental cyst presenting with profound anaemia

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ABSTRACT

A male infant presented acutely with profound anaemia and abdominal distension. At laparotomy, a huge omental cyst was found. We reviewed our experience of this rare cause of abdominal pathology identifying five further cases since 2005, with a variety of presentations. The optimal treatment, complete resection, is associated with a low morbidity. The presentation with profound anaemia and shock due to haemorrhage into the cyst is unusual.

key words: Anaemia, haemorrhage, mesenteric cyst, omental cyst, paediatric

INTRODUCTION

The reported incidence of mesenteric and omental cysts is 1 in 20,000 admissions to a paediatric hospital.[1]

There have been several reports in the literature describing the many ways these cysts can present; however, presentation with anaemia and shock requiring transfusion is a rare presentation reported in two out of 15 cases by Chung *et al*.[1] and a Danish case report of a female infant aged 14 months at presentation. She had haemoglobin of 2 mg/dL and ultrasound findings of a unilocular cyst. At laparotomy, there was an omental cyst containing 1-1.5 L of haemorrhagic fluid that was extirpated.[2]

MATERIALS AND METHODS

After the presentation of the most recent case described below, we undertook a review of our unit's experience of the management of this rare abdominal pathology. Retrospective case note review was performed on all children who presented to our institution with omental and mesenteric cysts between 2005 and the present day. Ethical approval was obtained from our local ethics committee before proceeding with the study. Information on demographics, mode of presentation and clinical course was extracted.

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CASE REPORT

A two-year-old boy presented with a three-day history of abdominal distension, pallor and lethargy. There were no associated gastro-intestinal symptoms and no history of trauma. There were no significant past medical problems. Until this acute episode, he was entirely well, born on the 25th centile but thriving and growing on the 50th centile.

On examination, he was pale and quiet. There was no jaundice; he was apyrexial with a tachycardia of 140 bpm. His blood pressure was 113/74 mm Hg; he was cool peripherally with a prolonged capillary refill. His oxygen saturation was at 100% in room air and had no signs of respiratory distress. His neurological examination was unremarkable. His abdomen was distended and on palpation, was soft although generally tender but without guarding or peritonism.

He had a normochromic normocytic anaemia with serum haemoglobin of 3.2 g/dL. The coagulation screen, liver function tests, renal function, electrolytes and lactate dehydrogenase were all within normal limits. His erythrocyte sedimentation rate was mildly elevated at 17 mm/hr. A Mantoux test was performed, which was negative. His rapid HIV PCR was equivocal and later confirmed negative.

An abdominal radiograph showed a paucity of bowel gas pattern generally. An ill-defined opacity appeared to be displacing small bowel loops to the right upper

quadrant [Figure 1]. An ultrasound scan demonstrated a multi-septated fluid-filled mass extending from the epigastrium to the pelvis with echogenic debris within. All solid organs were visualized and appeared normal [Figure 2]. He was resuscitated and received a transfusion of blood prior to surgery.

At laparotomy, he was found to have a large multi-lobed cyst filling the entire abdominal cavity containing more than two litres of haemorrhagic fluid [Figure 3]. The cyst appeared to be arising from the splenic hilum with a small element extending behind the spleen. The cyst was dissected free from surrounding structures and excised intact. There were no other abnormalities noted at laparotomy. He made an unremarkable recovery and has had no reoccurrence in the three months since surgery.

The multi-loculated cyst was 17 cm in its widest diameter containing haemorrhagic fluid. On histolopathologic examination, variably sized endothelial-lined vascular channels separated by fibro-adipose tissue with smooth muscle and lymphoid aggregates were noted [Figure 4]. These pathological features were consistent with a diagnosis of lymphangioma.

RESULTS

A review of patients presenting with mesenteric and omental cysts seen in this unit since 2005 identified further five cases. The male to female ratio was 1:2. The age range for this cohort was 2-10 years. The mean age at diagnosis was 4.5 years. However, at the upper end of age range was a 10-year-old boy. He had presented at the age of four years for investigation of "ascites". He was investigated, and no cause was found, but his "ascites" resolved without the need for intervention. He was discharged from follow-up and was entirely asymptomatic in the intervening period before his



Figure 1: Abdominal radiograph: General paucity of gas shadows. No calcification or pneumoperitoneum

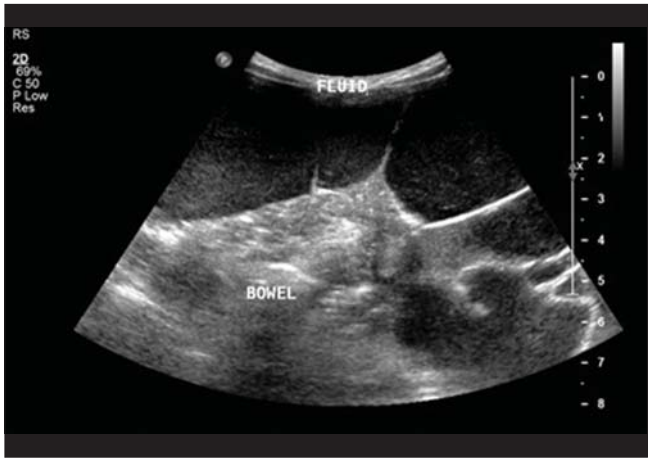


Figure 2: Ultrasonography: Multiseptated fluid collections from epigastrium to pelvis with displacement of bowel loops

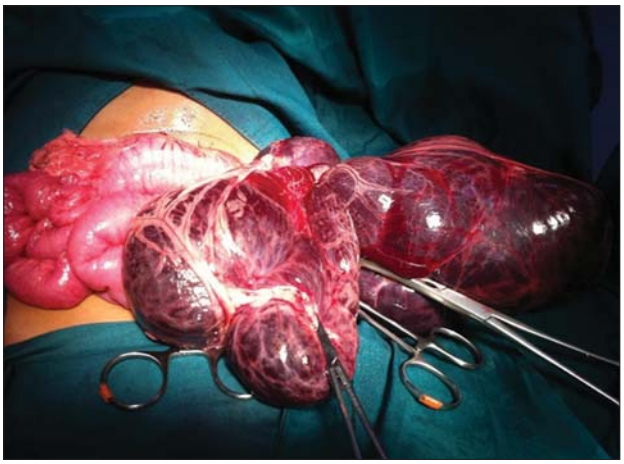


Figure 3: Intra-operative picture showing multilobulated cyst with normal transverse colon and small bowel displaced superiorly

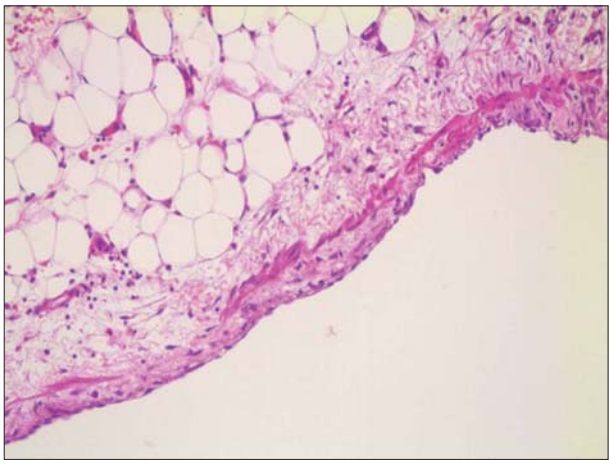


Figure 4: High power histology image showing an endothelial lining with adjacent smooth muscle and adipose tissue (Haematoxylin and eosin stain)

admission with an acute abdomen six years later when he presented with a two-day history of abdominal pain, vomiting and anorexia. He was pyrexial on examination and had lower abdominal tenderness (maximal left iliac fossa) with raised inflammatory markers (C-reactive protein 181 mg/L, WCC 36.2 × 10^9/L and erythrocyte sedimentation rate of 60 mm/hr). The other modes of presentation are summarized in Table 1.

We defined acute admission as children who had three or less days of symptoms. Four out of six patients presented in an acute manner. In three cases, the children had an acute abdomen (one with small bowel obstruction) and a presumptive diagnosis of appendicitis prior to surgery. The fourth acute presentation is the case presented above.

Two patients presented with chronic symptoms, both with at least a year's duration. One child had chronic abdominal pain with a shorter history of distension. The second child had a one-year history of distension with ascites of unknown cause.

The majority of patients had pre-operative radiological imaging with all but one patient having an ultrasound examination of the abdomen. One of the patients with an acute abdomen and small bowel obstruction proceeded directly to laparotomy with no radiological investigation. He was found at laparotomy to have a mesenteric cyst, which was a dumbbell shape draped around the terminal ileum. The cyst was removed without the need for bowel resection. One patient had cross-sectional imaging with a computerized tomography of the abdomen.

Minimally invasive surgical techniques were utilized in four patients. In three patients, laparoscopy was used as an additional diagnostic aid and in the fourth case for diagnosis and resection. Three of the cysts were present in the mesentery and three in the omentum. One patient required a limited bowel resection of 13.5 cm ileum due to the intimate relationship of the cyst to the bowel. Three of the cysts were multiple, and three were single. Five of the cysts were multi-locular and were filled with haemorrhagic fluid. The size of cysts ranged from 8 cm to 30 cm in the widest diameter. On histological examination, five of the six cases were considered to be lymphangiomatous in origin. There was one case of a mesothelial cyst. Post-operative in-patient stay ranged from two days for the laparoscopic case to six days with a mean of four days.

There were no cases of recurrence and one complication. The complication was a child who presented three months after resection with adhesive small bowel obstruction, which required open adhesiolysis. The follow-up period ranged from two months to 70 months with a mean follow-up of 28 months.

DISCUSSION

A mesenteric cyst was first reported in 1507 by Benevieni, a Florentine anatomist after an autopsy on an eight-year-old child.[3] Subsequently, in the nineteenth century, von Rokitansky[4] reported the first case of a chylous mesenteric cyst in 1842, and Gairdner described the first omental cyst in 1852.[5] The first report of surgical treatment of a mesenteric cystic mass was in 1880 by a French surgeon Tillaux.[6] A classification system was devised for mesenteric and omental cysts. They have been classified into four groups; embryological and developmental, traumatic or acquired, neoplastic and infective or degenerative.[7]

The mean age at the time of diagnosis is four years.[8] There is a male predominance in the paediatric population with a male to female ratio of 3:2.[9]

Lymphangiomas are the commonest cause of mesenteric and omental cysts comprising 90% of cases. Lymphangiomas are thought to arise from benign proliferation of heterotopic lymphatics that lack communication to the rest of the lymphatic system. Animal models do not support an obstructed lymphatics theory as where obstruction of the lymphatics is replicated, intra-abdominal cysts do not develop. Lymphangiomas are characterized by endothelial lined channels with smooth muscle and lymphoid aggregates

Table 1: Table summarising the case series

Case	Year	Sex	Age (yrs)	Presentation	Histology	Post op stay (days)	Complications	F/U Months
1	2005	M	10	Acute*	L	3	N	69
2	2005	F	3	Chronic	L	2	N	63
3	2008	F	5	Acute	L	6	N	38
4	2010	F	3	Chronic	M	4	N	17
5	2010	F	4	Acute	L	5	Y	14
6	2011	M	2	Acute	L	6	N	3

*Previous history of "ascites" 4 years old L- lymphangioma, M- mesothelial cyst

in the walls. The majority of the remaining cysts are mesothelial cysts. These are lined by mesothelium and are most commonly uni-locular with serous fluid within. These are thought to arise due to incomplete fusion of mesothelial leaves of the omentum.

Mesenteric cysts present with a variety of clinical presentations, both acute and chronic. In a review consisting of the summary of several case series of 281 children, the commonest symptom was abdominal pain in 40%.[10] This is consistent with other reviews that found 50%[8] to 58%[11] presenting with abdominal pain. Other common presentations include abdominal distension[12] and mass or bowel obstruction.[13] There are reports of cases of small bowel volvulus[14] and perforation complicating the cysts. Malignant transformation is rarely seen in adult patients.[11] Of interest is the intermittent "ascites" in one of our cases.

Anaemia is an uncommon presentation with one case of chronic anaemia due to an omental cyst requiring transfusions,[15] and three acute cases in a series of 15 cases from the St. Jude Hospital, Montreal between 1970 and 1990 being reported. Two of these patients required transfusion. This was the presenting feature in our index case.

In the majority of cases, a pre-operative diagnosis is not made. In all reviews, ultrasonography is reported as the best modality for diagnosis.[16] The sonographic features are fluid-filled, cystic multi-septated masses with lobules that may be anechoic or contain internal echoes or sedimentation due to debris.[17] On computed tomography, there may be no discernible mesenteric wall, and the cyst may have the characteristics of fat if the contents are chylous.[18] Laparoscopy is emerging as an additional diagnostic tool, which can be utilized for resection in some cases as in one of our cases.[19,20]

Complete surgical resection has an excellent prognosis. The reported recurrence rate in a group of 162 adults and children was 6.2%.[11] Recurrence is more likely in cases with incomplete excision or with retroperitoneal extension.[11] The mortality from the above series was reported as 2%.

There has only been one reported mortality in the paediatric literature. The case was a 23-month-old child who had small bowel volvulus associated with a mesenteric cyst and mesenteric defect.[21]

Omental and mesenteric cysts are rare causes of abdominal pathology, and the treatment of choice is complete resection, which was possible in all cases from this unit, requiring a limited small bowel resection in one case. This is associated with a low morbidity and mortality. The experience of this unit mirrors the varied modes of presentation reported in the literature. However, acute presentation with anaemia requiring pre-operative transfusion is a rare presentation.

ACKNOWLEDGMENT

The Dowager Countess Eleanor Peel trust for awarding the Peel Travelling Grant towards the funding of the first author's fellowship year.

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Cite this article as: CAdikibi BT, Wood R, Pillay K, Millar AJ. Omental cyst presenting with profound anaemia. Afr J Paediatr Surg 2013;10:180-4.

Source of Support: None. **Conflict of Interest:** None declared.