Case Report

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Neonatal abdominal cocoon arising from atypical sequelae of intestinal malrotation: A case report

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Ladd's procedure,
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ABSTRACT

Background: Abdominal cocoon (AC) is the partial or complete encasement of the intestines and sometimes other abdominal organs by a fibro-collagenous sac causing varied presentations of intestinal obstruction. It has been found in all age groups from neonates to the elderly although neonatal AC is quite rare and only very few cases have been reported. In neonates, the presentation could mimic other causes of partial or complete intestinal obstruction and preoperative diagnosis is usually difficult.

Case Presentation: We report an atypical sequela of intestinal malrotation causing neonatal intestinal obstruction due to omental encasement of the small and large bowel. The diagnosis was made intraoperatively though contrast gastrointestinal series suggested the partial obstruction at the duodenal-jejunal level. The patient made a good postoperative recovery after extraction of the bowel from the cocoon (omental sac), partial omentectomy, and Ladd's procedure.

Conclusion: A neonatal abdominal cocoon caused by an omental encasement in a malrotated intestine is a unique presentation and a rare cause of neonatal intestinal obstruction.

INTRODUCTION

Neonatal intestinal obstruction (NIO) is one of the commonest emergency presentations to the neonatal surgeon. [1-3] The common causes include intestinal atresia, malrotation, anorectal malformation, and Hirschsprung's disease. [3] Preoperative diagnosis may be quite difficult, however, prompt diagnosis and immediate intervention are usually required to curtail accompanying morbidity and mortality. [3]

Abdominal cocoon (AC) is a very rare cause of NIO and most neonatal surgeons have not come across this in their practice as only a few have been reported in the literature. [4] Although it is also seen across all other ages, neonatal AC (NAC) presents a unique set of aetiologic, diagnostic, and treatment peculiarities. [4]

The abdominal cocoon first coined by Foo [5] refers to the entrapment of the bowel within a sac, and this forms the basis of its clinical presentation. Typically, it affects the mobile small bowel alone, however, it can also involve the large bowel and other abdominal viscera. It has generally been classified as primary or secondary; congenital or acquired based on its etiology. [6-8] More cases have been reported in adolescents and adults with a few reports from Nigeria. [9]

For neonatal surgeons especially those in resource-poor settings, having this addition to the list of differentials for NIO diagnoses can be very helpful. We report a unique case of NAC and review the literature on the abdominal cocoon.

CASE REPORT

A 5-day-old term female neonate was admitted via the NICU with persistent non-bilious, non-projectile, postprandial vomiting which started a few hours after delivery. A low-grade continuous fever suspected to be due to sepsis was noticed a day after delivery accompanied by mild abdominal distension. Meconium was passed within 24 hours.

Pregnancy, labor, and delivery were uneventful. There was no history of polyhydramnios present. Examination showed a pink, febrile, tachycardic

(heart rate 152-156), dehydrated, and anicteric baby. Her birth weight was 2.4kg. Examination showed a soft, non-tender, and mildly distended abdomen, soft, with a normally sited patent anus. The rest of the examination was grossly normal.

The baby was resuscitated and placed on IV fluids and antibiotics. Nasogastric tube (NGT) drainage was bilious. The hematological profile showed leukocytosis from sepsis and blood chemistry showed high urea and creatinine levels due to dehydration and acute kidney injury.

Plain abdominal x-rays showed a grossly dilated stomach and paucity of gas in the distal bowel (Fig. 1A). Abdominal distension resolved after 2 days while NGT drainage became clear and the baby started passing stools. He was allowed dextrose water via NGT which was tolerated for 5 days before the residual became bilious again.

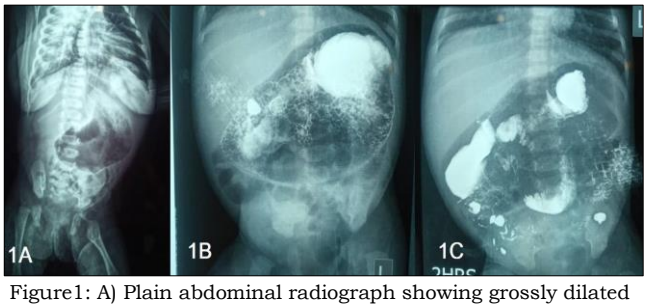
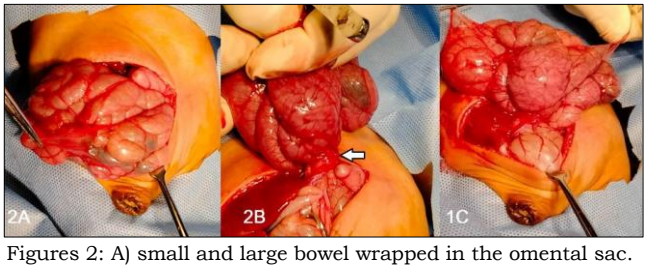


Figure1: A) Plain abdominal radiograph showing grossly dilated stomach and paucity of gas in the distal bowel, B and C) Barium meal and follow-through showing dilated stomach and duodenum and jejunum.

The upper GI series suggested dilated stomach and proximal duodenum up to the third part; there was delayed passage of minimal contrast distally (Fig. 1B,1C). The abdominal ultrasound was unremarkable due to the gaseous distension.

At exploratory laparotomy on the 19th day of life, the whole small bowel was enclosed in a thin membrane formed by the greater omentum, the neck of the sac was narrow entrapping the afferent and efferent loops of the small bowel which traversed it (Fig. 2A, 2B). The whole large bowel was located on the right, peritonealized, and enclosed in the same sac lying parallel to the small bowel which was lying anterolaterally on the left (Fig. 2C). The sigmoid colon runs straight down to the rectum in the midline without forming its loop. The stomach and duodenum were dilated and the duodenojejunal junction was in the normal position to the left of the vertebra, the caecum was below the liver towards the epigastrium. Other viscera were normal in appearance and position. The whole small bowel was extracted through the neck of the sac and there were no interloop adhesions, but bands were holding the mesentery together. The Sac, which was the omentum, was formed by the parallel lying ascending and descending colon with the transverse colon at the summit. The peritoneum over the large bowel was released to separate the colon which had narrow mesocolon and Ladd's procedure was done for the small bowel which also had narrow mesentery. Appendectomy was then done to prevent future diagnostic dilemmas.



Figures 2: A) small and large bowel wrapped in the omental sac. B) Narrow neck of the sac (arrow) through which the bowel was extracted. C) Relative position of the small and large bowel in the sac.

The immediate postoperative period was satisfactory. The patient moved the bowel on the second postoperative day and oral intake commenced on the third day. The patient was discharged in good condition.

DISCUSSION

Cleland [10] in 1868, first described peritoneal encapsulation as an embryologic developmental anomaly in which there is an accessory peritoneal membrane, believed to be derived from the yolk sac. This wraps up the small bowel separating it from the parietal peritoneum and the omentum. This was described as an abnormal developmental process and is typically asymptomatic and not associated with inflammation. [6,7] There is a total or partial encapsulation of the bowel by a thick fibro-collagenous membrane termed peritonitis Chronica fibrosa incapsulata. [4,5] Other names that have been used to describe it include sclerosing peritonitis, encapsulating sclerosing peritonitis, and chronic encapsulating fibrosis. Most of these terminologies suggest an inflammatory component to this pathology, which isn't always so. This has led to some confusion and controversies in the literature. [5]

AC has generally been divided into the primary or idiopathic and the secondary types. The primary type has no known cause, but the encasing peritoneum is believed to be derived from the yolk sac, mesocolon, and possibly the omentum. [4]

Secondary AC is due to a known cause, usually a preceding local or systemic inflammation from causes such as meconium peritonitis, prolonged peritoneal dialysis, ventriculoperitoneal shunts, bacterial or tuberculous infection of the peritoneum, medications such as beta-blockers, methotrexate, and exposure to toxic substances. [11] Neonates presenting within the first few days of life may have a congenital AC which

could be idiopathic from no known cause, or secondary to an antenatal inflammatory event such as meconium peritonitis. [4]

In this case, the cocoon was formed by the greater omentum, which has not been described before in English literature. It was a thin vascular membrane that folded on itself and formed a cord-like tissue at the neck of the sac, there were no adhesions found suggesting the absence of preceding inflammatory events.

Wei et al [11] reported that AC can be categorized into 3 types according to the extent of the encasing membrane, in type 1—the sac partially encapsulates the intestine; in type 2—the whole intestine is encapsulated by the membrane, and in type 3—the whole intestine and other intraperitoneal organs are encapsulated by the membrane. [7] Symptomatology may depend on the extent of the entrapment. Our case fits into the Wei type 2 because the small bowel was completely encapsulated and the large bowel which formed the posterolateral wall was also peritonealized.

The patient may be asymptomatic, and the cocoon may be found as an incidental finding, depending on the extent of the encasement. [5,9] However it can also present with the acute obstruction which can be complete or partial. [9] In some cases, the recurrent subacute obstruction may also be noted as seen in our patient who had a partial obstruction as she was still passing stool minimally. An adequate history is usually very important to exhaust all other differential diagnoses such as internal hernias, malrotation, and volvulus.

Mesocolic hernias are rare congenital lesions that result from abnormal rotation of the gut in which the small intestine herniates posteriorly to the mesocolon. [12] This is not so in our case as the small bowel was encased in the omentum and was anterior to the large bowel loop.

Imaging studies are mostly inconclusive. [9] Plain abdominal x-rays may be unremarkable, or it may also show dilated stomach and proximal bowel, there may be a paucity of gas in the distal bowel, or crowding of small bowel gas shadows. [5,7] If there are no signs of complete obstruction, contrast gastrointestinal x-ray series may show delayed intestinal transit time. However, no definite transition point is usually seen. Abdominal ultrasound has been reported to show a cauliflower sign depicting the encased bowel on a stalk of its narrow mesentery base. This is usually obscured if there is proximal dilatation and excess intraluminal gas but may be more prominent in the presence of ascites. [7,11] In adults, a contrast-enhanced CT scan may be the best option for investigating suspicion of an abdominal cocoon but the risk of exposure to such high doses of radiation, makes it unattractive in neonates.

The treatment is usually the removal of the encasing membrane and adhesiolysis with care not to injure the bowel. [4, 5, 11] The whole bowel should be inspected for ischemia, prenatal pathologies, or injuries that may necessitate resection and anastomosis, although this is not usually required in most cases.

The index case had an extraction of the whole bowel through the narrow neck of the sac. The minimal peritoneal release was done for the large bowel that formed a knuckle though there were no adhesions between the sac and small bowel. Usually, the prognosis is excellent if there is no associated bowel gangrene, and a careful surgery is done. [11]

In conclusion, an omental cocoon is a rare cause of neonatal intestinal obstruction. A high index of suspicion is required for the diagnosis and surgeons should be aware of its existence, presentation, and treatment. Prognosis is excellent after correct diagnosis and intervention.

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