

## THE ABDOMINAL COCOON – A REPORT OF THREE CASES AND REVIEW OF LITERATURE

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### ABSTRACT

The abdominal cocoon is a rare cause of intestinal obstruction in adolescent girls. Since the first reported case in 1978, thirteen other cases have been reported. We present three cases of abdominal cocoon treated in this hospital during the past 10 years including two who presented successively in one year. The pathology of this condition is discussed and a comprehensive literature review is made. Treatment modalities are also covered.

**KEY WORDS:** *Abdominal cocoon, small intestinal obstruction, adolescent girls.*

### INTRODUCTION

The abdominal cocoon is a rare cause of intestinal obstruction in adolescent girls. It is characterized by a fibro-collagenous encasement of either part or the whole of the small intestine, resulting in features of intestinal obstruction.

Fourteen cases have been reported in the literature by 1991 in females aged between 4 and 18 years. All have lived in subtropical or tropical environment<sup>1-5</sup>. We present three cases of abdominal cocoon in adolescent girls aged 10, 12 and 18 years respectively. Two cases presented with recurrent features of intestinal obstruction and the third case presented with lower abdominal mass associated with some discomfort.

Laparotomy revealed encasement of the small intestine from the duodenojejunal flexure to the ileo-caecal junction by fibro-collagenous peritoneal adhesions. The patients had adhesiolysis and post-operative recovery was uneventful. A review of the literature and management of this unusual condition is highlighted.

### Case Reports

#### Case 1.

O.O., an 18 year old female patient presented on 16th October, 1998 with recurrent lower abdominal pains of 5 months duration. Pain was sharp and colicky, non-radiating and relieved by analgesics. She had nausea but no vomiting. Her menarche was at 15 years and last menstrual period was 26th September, 1998, and lasted 3 days.

Examination revealed lower abdominal tenderness, maximal in the left iliac fossa. Pelvic ultrasound scan showed a left pelvic collection with mixed echogenicity. Laparotomy on 21st October, 1998 showed dense generalized membranous adhesions encasing the whole of the small intestine from the duodenojejunal junction to the ileo-caecal junction. Figures I and 2 show the loops of small intestine encased in membranous adhesions. A cystic

collection containing serous fluid was found in the region of the left ovary, extending to the left paracolic gutter. Adhesiolysis was done and a long pelvic appendix involved in the adhesions was removed. The left fallopian tube and uterus were identified but the right fallopian tube was walled off in the pelvis.

She had an uneventful post-operative recovery and was discharged home on the 14th post-operation day. She has remained well and is being followed up in the outpatient clinic.



Fig. 1: Loops of small intestine encased in membranous adhesions



Fig. 2: Loops of small intestine after adhesiolysis

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**Case 2.**

U.S, a 12 year old female patient presented on 4th November, 1998 with recurrent abdominal pains of one week duration and abdominal swelling of three days duration. The pain was colicky, intermittent, non-radiating with no known relieving or aggravating factors. The swelling was periumbilical and patient could not remark about variation in size. Her menarche was two months prior to presentation.

Examination revealed a healthy looking young girl, not pale, anicteric and febrile. The abdomen revealed a uniform, round periumbilical mass, not tender, firm, smooth with limited mobility.

The percussion note was dull and bowel sounds were normal. Rectal examination was normal with well formed stool on examining finger. The provisional diagnosis was a mesenteric

cyst, the differential diagnoses were abdominal tuberculosis and abdominal lymphoma. Plain abdominal x-ray showed soft tissue mass around the central abdomen with gas in the rectum. Abdominal ultrasound scan showed distended bowel loops marked in the right iliac fossa with significant intraperitoneal fluid collection.

She had exploratory laparotomy on the third day of admission. Findings include multiple loculated cysts containing serous fluid, small bowel encased in fibrous peritoneal adhesions from the duodenojejunal flexure to the terminal 2-5 centimetres of ileum. The rest of the abdominal organs were normal. She had adhesiolysis and incidental appendectomy. Her post-operative recovery was uneventful and she was discharged home on the 10th post-operative day. She has remained well so far.

*Table 1: Reported cases of abdominal cocoon*

Case no and reference	Age(year)	Symptoms	Abdominal observations	Operative findings	Procedure
1.1	14	Abdoinal pain vomiting	Mass in right iliac fossa	Cocoon is distal third of small bowel	Resection (limited hemicolectomy)
2.1	14	Abdomina pain vomitng	Mass in hypogastrium	Cocoon of small bowel, liver adhesion	Lysis, Noble's plication appendicetomy
3.1	15	Abdomina pain vomitng, distension	No mass	Pelvic adhesions of distal third of small bowel	Lysis decompression, ileotransverse colostomy
4.1	16	Abdomina pain vomitng, mass	Mass in hypogeastrium	Cocoon of whole of small bowel	Lysis and appendicectomy
5.1	17	Abdomina pain, vomitng, distension	No mass	Cocoon of distal half of small bowel, pelvic adhesions	Lysis and appendicectomy
6.1	15	Abdomina pain, vomitng, distension, constipation	No mass	Cocoon in distal quarter of small bowel	Lysis and lymph node biopsy
7.1	16	Abdomina pain vomitng, distension, constipation	No mass	Cocoon of whole of small bowel	Lysis and appendicectomy
8.1	14	Abdomina pain, mass	Periumbilical mass	Cocoon of whole of small bowel	Lysis and appendicectomy
9.1	13	Abdomina pain vomitng, mass	Mass in hypogeastrium	Cocoon of whole of small bowel, generalised adhesions	Lysis and appendicectomy
10.1	18	Abdomina pain, mass	Mass in hypogeastrium	Cocoon of whole of small bowel	Lysis and appendicectomy
11.8	4	Symptoms of obstruction	Mass in abdomen	-----	-----
12.7	17	Abdomina pain	Mass in hypogeastrium	Cocoon of distal ileum	Lysis
13.9	14	Vomiting, distension	None	Cocoon of whole of small bowel	Lysis
14.10	15	Abdominal pain, vomiting	Central abdominal mass	Cocoon in distal two third of small bowel	Lysis lymph node biopsy appendicetomy

### Case 3.

O.A., a 10 year old girl, one of identical twins and 4th of 8 siblings, presented on 2nd March, 1991 with history of progressive weight loss and lower abdominal discomfort of two months duration. She had good appetite and normal bowel action. Five months prior to these symptoms, the girl had suffered a generalised abdominal pain and distension and was treated in a private hospital with antibiotics. She got better and had remained well until the last 2 months.

On examination she appeared well. The abdomen was soft and non-tender. There was a mass in the lower part of the abdomen occupying the left iliac fossa and extending into the pelvis. It was firm, non-tender and not mobile. On rectal examination a mass was felt in the pelvis. The provisional diagnosis was ovarian tumour; differential diagnoses considered were uterine mass and Burkitt's tumour in the pelvis. Plain abdominal x-ray showed soft tissue opacity displacing the descending colon laterally. Intravenous urography showed normal function of both kidneys with normal pelvi-calyceal system. Left ureter was slightly displaced laterally by a mass thought to be retroperitoneal.

Laparotomy revealed extensive fibrous adhesions between all the adjacent surfaces of all abdominal viscera. A loop of small intestine was adherent to the parietal peritoneum of the anterior abdominal wall. The small intestine was enclosed in a ball of dense fibrous adhesion in the left iliac fossa and bound down to the pelvic viscera and the posterior abdominal wall. This constituted what had been clinically felt as the abdominal mass. The pelvic viscera were also involved in fibrous adhesions. Adhesiolysis was done using a combination of blunt and sharp dissection, care being taken to avoid injury to the loops of small intestine. Appendicectomy was also carried out. Post-operatively she was treated with intravenous fluids and parenteral nutrition solutions. The recovery was uneventful and within 3 weeks after operation she regained over 4kg of her body weight and was discharged home. She has remained well ever since.

### **PATHOLOGY**

In all three cases specimens of peritoneal fluid, lymph nodes and adhesive bands were sent for cytology and pathological analysis. Peritoneal fluid cultures grew no organism and were negative for acid and alcohol fast bacilli in the three patients. Histology of biopsies from lymph nodes and adhesive bands showed only non-specific inflammatory changes and varying amount of fibroblastic proliferation. The appendix was reported normal in two of the patients (cases one and two) and congested only, in the third case. The findings in all the three cases were suggestive of primary non-specific inflammatory pathology.

### **DISCUSSION**

Small intestinal obstruction is a common surgical emergency. Previous abdominal surgery is the commonest cause of peritoneal adhesions and fibrosis. Other important aetiological factors include infections such as tuberculosis and peritonitis from perforated viscus. Other rare causes of peritoneal fibrosis include sclerosing peritonitis due to the  $\beta$ -adrenergic drug practolol<sup>6,7</sup>, peritoneal mesothelioma or fibrous reaction to asbestos<sup>8</sup>, mesenteric panniculitis<sup>9</sup> and the carcinoid syndrome<sup>10</sup>.

The patients reported in this series did not have previous abdominal surgery and so their condition cannot be ascribed to post-operative adhesion and fibrosis. There was no evidence to suggest tuberculous peritonitis, there were no tubercles in the peritoneal cavity and histology of the biopsied fibrous tissue showed no Langhan's giant cells. The peritoneal fluid aspirate contained no acid - and alcohol-fast bacilli. There was no evidence of perforated viscus as the appendix, small intestine, gallbladder and the duodenum did not show any evidence of perforation. Peritoneal mesothelioma was excluded by lack of a history of contact with asbestos and by the pathological findings. A diffuse fibrosis of the peritoneal cavity is occasionally seen in patients with the carcinoid syndrome; though the precise cause is unknown, histology excluded this disorder. Mesenteric panniculitis is a rare disorder affecting adipose tissue in the small bowel mesentery and histology also excluded this disorder. There is no history of ingestion of the  $\beta$ -adrenergic drug practolol in any of these patients.

We believe that these adolescent girls seem most likely to have had the condition described as the abdominal cocoon. This condition was first reported by Foo et al<sup>6</sup> in 1978 in ten adolescent girls from Singapore who presented with features of small intestinal obstruction. At operation the small intestine was either totally or partially coiled up in dense adhesions extending to surrounding structures. In 1980, Marinho and Adelusi<sup>2</sup> reported a similar case in a 17 year old adolescent girl from Ibadan. In all, 14 cases<sup>1-5</sup> have been reported in the world literature (Table 1). All previous reports have been in females between 4 and 18 years old and all have lived in subtropical or tropical areas. All have presented with acute or subacute intestinal obstruction, in five cases there was abdominal distension and in nine cases an abdominal mass was present as shown in table 1.

The aetiology of this uncommon condition is unknown but the commonest theory is one of membrane formation secondary to peritonitis. Retrograde menstruation, particularly if associated with a virus infection, could cause peritonitis. The age and sex distribution of the patients described supports this theory<sup>1</sup>.

Alternatively, it should be noted that the 'cocoon' is often separate from the parietal peritoneum. The 'cocoon' may represent a congenital abnormality which comes to light in girls who have retrograde menstruation causing peritonitis<sup>2</sup>. The consistent siting of the lesion with invariable involvement of the ileo-caecal junction also supports the idea of a developmental abnormality.

The treatment of this condition is lysis of the encasing membrane at laparotomy by careful dissection and excision. In most cases, incidental appendectomy has also been performed as in our cases. Although the prognosis of the condition appears to be excellent, it is difficult to predict the effect on the reproductive health of the patients in later life.

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