

Ileocecal Knotting Causing Intestinal Obstruction on Early Postpartum Period: Case Report

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Introduction: Intestinal knotting syndromes are rare causes of mechanical bowel obstruction, ileo-sigmoid knotting being the most common type. Ileocecal knotting is the rarest type among knot syndromes and there are few case reports across the world.

Case Presentation: We present a case of ileocecal knotting in a postpartum patient with abdominal distention, vomiting and failure to pass feces and flatus. Plain abdominal radiograph revealed dilated small bowel loops with multiple air fluid levels. Exploratory laparotomy was performed with intra-operative finding of ileocecal knotting with viable bowel, mobile cecum and ascending colon and gangrenous tip of appendix. We untied the knot and performed an appendectomy. Due to intra-operative instability of the patient's vital signs, right hemicolectomy was differed and cecum as well as ascending colon were fixed to right posterolateral abdominal wall.

Clinical Discussion: Ileocecal knotting is a very rare finding, and few case reports have been published. The intra-operative findings of the mobile cecum and ascending colon are consistent with previous reports and it is a predisposing factor for knot besides low BMI and young age of our patient. Surgical management is dictated by intra-operative conditions such as viability of the bowel, intra-operative stability of the patient, extent of resection and length of the remaining bowel. Surgical options include en bloc resection of knot with gangrenous bowel and end to end anastomosis or end ileostomy with closure of distal end.

Conclusion: Ileocecal knotting should be considered as a differential diagnosis for patients presenting with intestinal obstruction with unusual radiographic findings. Early diagnosis and prompt surgical intervention prevents bowel infarction and enhance the prognosis. Preoperative abdominal CT scan is helpful in such cases with unusual radiographic findings.

Keywords: ileocecal knotting, bowel obstruction, knotting, acute abdomen, case report

Introduction

Background

Mechanical bowel obstruction can be caused by extrinsic, intrinsic or intraluminal causes.¹ Intestinal knotting is a rare but life-threatening cause of both small and large bowel obstruction resulting in a closed double-loop obstruction.² Different types of intestinal knot syndromes have been discussed across various literatures, that include ileo-sigmoid, ileo-ileal, appendico-ileal and ileocecal knotting. The commonest being ileo-sigmoid knotting.¹⁻⁴

Complete bowel obstruction leads to dilation of the bowel proximal to the site of obstruction, with accumulation of gas and intestinal contents and collapsed bowel distal to the site of obstruction.^{1,3,5} With an increase in the duration of obstruction, bowel distension gradually worsens leading to a higher risk of compromised blood perfusion to the bowel, resulting in necrosis or perforation. The occurrence of these conditions increases morbidity and mortality.³ When intestinal knotting occurs, the risk of strangulation will be high in both the active and passive components of the knot and it is associated with poor prognosis unless intervened early.¹

Pre-operative diagnosis of intestinal knotting as a cause for intestinal obstruction is very challenging because radiograph shows dilated loops of both the large and small bowels.^{1,3,5-7} Ileocecal knotting causing acute bowel obstruction occurs when a loop of ileum encircles around the cecum and ascending colon. It needs a rapid and prompt resuscitation followed by surgical

intervention.^{1,2,8–10} Surgical management requires resection of gangrenous bowel segments with end-to-end anastomosis or temporary diversion with stoma formation if intra-operative patient and bowel conditions are not conducive for anastomosis. Intra-operative finding of viable ileocecal knotting requires right hemi-colectomy and ileo-transverse anastomosis.^{1,2}

In this report, we present the case of 20-year-old female, para I patient on her first postpartum day after she gave birth through caesarian section (C/S) at St. Paul's Hospital Millennium Medical College (SPHMMC). The patient was in critical condition having signs and symptoms of acute abdomen secondary to bowel obstruction and subsequently an exploratory laparotomy was performed with intra-operative finding of viable ileocecal knotting with mobile cecum and ascending colon.

Based on our literature search, so far, there was only one reported case of ileocecal knotting from Ethiopia and there are very few cases reported across the world, although none of them occurred during the postpartum period. In this case report, we present a case of ileocecal knotting as a rare cause of intestinal obstruction during the early postpartum period, adhering to the SCARE criteria.¹¹

Case Presentation

A 20-year-old female patient presented with progressive abdominal distension associated with abdominal cramp, failure to pass flatus and feces as well as frequent vomiting of ingested and bilious matter of one-day duration. The day before, she gave birth through C/S for an indication of severe fetal bradycardia. She also had preeclampsia without severity signs and an adult onset malnutrition with a body mass index (BMI) of 16.5Kg/M². She had no history of previous abdominal surgery. She denied any similar complaint in the past or previous history of bowel habit change or constipation with passage of a blood mixed stool. She did not have any known chronic medical illness.

On physical examination she was acutely sick looking and in pain. Her blood pressure was 90/70mmHg, pulse rate was in the range of 116–120 beats per minute, respiratory rate was in the range of 22–24 breaths per minute. She did not have an objective fever record. Her abdomen was grossly distended and symmetric with visible peristaltic bowel movements. There was diffuse direct tenderness all over her abdomen, more at the right lower quadrant. There was a positive sign of intraperitoneal fluid collection with shifting dullness. There was a well approximated suprapubic surgical wound with clean dressing. Her digital rectal examination finding was unremarkable. She had a 3cm long, linear, vertically oriented, superficial wound on her left labia-majora which was initially thought to be an iodine burn to the skin, by the obstetrics team.

She was catheterized and resuscitated with two bags of normal saline and produced 200mL of urine over an hour. A nasogastric tube (NGT) was inserted but with no significant output. She was investigated with Complete blood count (CBC) and plain abdominal radiograph. Her white blood cell count was 1780 cells/ μ L with neutrophil predominance of 88%, hemoglobin of 12.7mg/dl and platelet count of $78 \times 10^3 / \mu$ L. Her renal function test was normal and her liver enzymes were slightly elevated above the normal range but was not significant. Her plain abdominal radiograph (Figure 1) shows centrally located multiple-air fluid levels with dilated peripherally located large bowel loop and whitening of the pelvic space indicating a fluid collection. Abdominal computed tomography (CT) scan was not requested because we work in a resource limited setting and for patients with bowel obstruction, we request CT scan when there is possibility of alternative diagnosis or when tumor obstruction is suspected.

After obtaining informed written consent, she was explored under general anesthesia with a diagnosis of acute abdomen secondary to mixed bowel obstruction secondary to small bowel volvulus. The intra-operative finding was a distal ileum encircling the cecum and proximal ascending colon, both of which were mobile and not attached to the right posterolateral abdominal wall. Both the ileum and cecum as well as ascending colon were viable (Figure 2). The tip of the appendix was entangled with the ileal knot, and it was gangrenous. The proximal small bowel and distal large bowel were grossly distended otherwise in their normal location. There was about four-liter of serous intraperitoneal fluid.

Intra-operatively, the patient was persistently hypotensive (80/50 mmHg) since after the induction of anesthesia and was started on vasopressor support. Considering the patient's instability and viability of the bowels involved in the knot, right hemicolectomy was differed. Proximal and distal decompression of the distended small and large bowels was

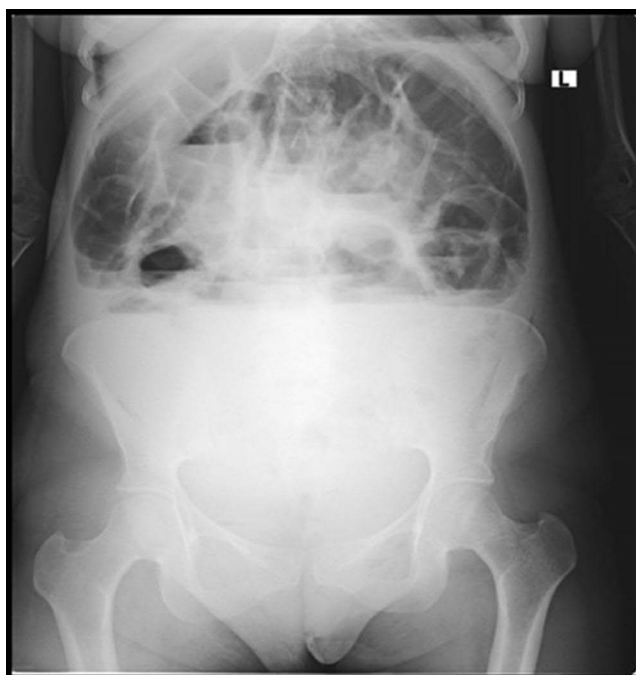


Figure 1 Plain erect abdominal radiograph of the patient. Multiple central air fluid levels suggesting dilated small bowel loops. Whitening of the lower abdomen and pelvic space suggesting intraperitoneal significant fluid. Significantly dilated large bowel loop.



Figure 2 Intraoperative picture of the ileocecal knotting. The distal ileum appeared dusky up on initial evaluation which later regained its normal color after the knot was released and warm pack.

performed respectively. Cecum and ascending colon were fixed to the right posterolateral abdominal wall using interrupted sero-muscular stitches with non-absorbable suture (Silk 2–0).

Post-procedure, she was extubated and kept nothing by mouth (NPO) for 24 hours and put on maintenance fluid with replacement of her loss. She was persistently hypotensive despite fluid resuscitation and vasopressor support and hence she was put on double antibiotics with Ceftriaxone 1gm, IV, BID and Metronidazole 500mg, IV, TID with consideration of septic shock of genitourinary focus. She gradually deteriorated and up on re-evaluation on her first post-operative day, the superficial

wound on her vulva has expanded upwards to the suprapubic area with clinical signs of Fournier's gangrene. Therefore, we revised her antibiotics to Meropenem 1gm, IV, BID, Vancomycin 1gm, IV, BID and Metronidazole 500mg, IV, TID. After informed written consent was obtained, she was taken back to the operation theatre for radical debridement of the Fournier's gangrene and subsequently transferred surgical intensive care unit (ICU) intubated due to persistent septic shock and delayed awakening from anesthesia. Unfortunately, she passed away with multi-organ failure due to refractory septic shock on her second post-operative day after the initial exploration.

Discussion

Mechanical obstruction is among the common causes of bowel obstruction in developing countries. A diagnosis of acute abdomen secondary to either small or large bowel obstruction accounts for a significant proportion of patients that undergo emergency laparotomy in Ethiopia.¹²

Intestinal knotting occurs when a loop of bowel is wrapped around a loop of another bowel segment forming a knot, the most common being ileo-sigmoid knotting.^{1,3,6,13,14} Other rare types of intestinal knot syndromes have been reported from different parts of the world, and they include ileo-ileal, ileocecal and appendico-ileal knotting.^{1,5,7-9} Ileocecal knotting is the rarest and worldwide there are few case reports published, including Ethiopia.^{1,2,10} Correct preoperative clinical diagnosis of these patients is made in less than 20% of cases.^{2,10} Preoperative utilization of abdominal CT scan might be suggestive of ileal knot syndromes with a finding of "whirl of mesentery" adjacent to the transition zone on the large bowel. Although, this finding on CT scan is not specific, it should raise suspicion of ileal knot syndromes.² However, in our case, we did not request abdominal CT scan and decision was made based on plain radiograph findings.

The intra-operative finding of our patient was a viable ileocecal knotting, with entangled and infarcted tip of the appendix, mobile cecum and ascending colon to the level of hepatic flexure as well as distended small and large bowel loops. Unlike other reports of this pathology, where most patients were found to have gangrenous bowel intra-operatively, our patient had viable ileum and cecum. This could be explained by the relatively early diagnosis of bowel obstruction and surgical intervention, while she was in-patient as she was on her first postpartum day after C/S delivery.

The intra-operative finding of mobile cecum and ascending colon is similar with other case reports, and it seems that it is the predisposing factor for the occurrence of the knot in addition to low BMI and young age of our patient.^{1,2,8,10}

Surgical management of ileocecal knotting is dictated by the intra-operative findings of presence or absence of gangrenous bowel, intra-operative condition of the patient, the extent of bowel resection performed and the length of the remaining viable bowel. Usually, en bloc resection of the knot with the gangrenous bowel and end to end/side anastomosis is done in a patient that is not in shock, minimal degree of bowel edema and minimal peritoneal contamination.^{1,2,5,7,10} On the other hand, end ileostomy with mucus fistula or closure of distal end is the preferred option in unstable patient who is in shock, with gangrenous obstruction and marked edema of the remnant viable bowel.^{1,2} In our patient, we untied the knot and performed an appendectomy with cecopexy (Suture fixation of cecum and ascending colon to the right posterolateral abdominal wall using non-absorbable suture). Considering the risk of future recurrence, the best treatment for this patient would have been right hemicolectomy and ileo-transverse anastomosis. However, the patient was very unstable intra-operatively as she was in shock and on vasopressor support.

Conclusion

Intestinal knot syndromes, particularly, ileocecal knotting should be considered as a differential diagnosis in patients with unusual radiographic findings and it is imperative that early diagnosis of bowel obstruction with thorough resuscitation followed by early surgical intervention will improve the post-operative outcome as well as decrease the morbidity and mortality of these patients. Preoperative abdominal CT scan should also be considered in such cases with an unusual finding on plain abdominal radiograph.

Ethical Approval

Informed written consent was obtained from the patient for publication of this case report before she passed away and ethical approval was also obtained from the Institutional Review Board (IRB) of St. Paul's Hospital Millennium Medical College with a Ref. No PML3/277.

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Disclosure

We declare no conflict of interest.

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