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Ileosigmoid knotting in patients under 25 years of age: A report of two cases[☆]



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ABSTRACT

INTRODUCTION: Ileosigmoid knotting is a rare cause of acute abdomen with high morbidity and mortality. Its diagnosis is infrequently made before surgery because of its varying ways of presentation and rarity. **PRESENTATION OF CASE:** The first was a 21-year-old male who presented with a history of sudden generalized abdominal pain and progressive abdominal distension. He was pale and severely dehydrated. His extremities were cold and clammy. His pulse rate was 110 per minute and blood pressure was 90/50 mmHg.

The second case was 20-year-old male who presented with similar symptoms as above. He was not pale but mildly dehydrated. His pulse rate was 92 per minute and blood pressure 110/70 mmHg.

Both patients were resuscitated and had exploratory laparotomy a few hours after presentation. The first patient was found to have ileosigmoid knotting with gangrenous sigmoid colon and terminal ileum. He had Hartmann's procedure and right hemicolectomy with ileo-transverse anastomosis.

The second patient was found to have ileosigmoid knotting with viable loops of bowel. He had careful detorsion, sigmoidectomy with primary anastomosis. Both patients' have good outcome.

DISCUSSION: This is to report two cases of ileosigmoid knotting in two male patients aged 21 and 20 years, respectively, with the hope of increasing awareness.

CONCLUSION: Ileosigmoid knotting though more common in fourth or fifth decade of life, can also occur in the 2nd decade. Early diagnosis, careful resuscitation and skilful surgical intervention will improve outcome.

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1. Introduction

Ileosigmoid knotting is rare cause of acute abdomen. It is more commonly seen in parts of Africa, Asia, and the Middle East but less so in other parts of the world like the UK where reports are sporadic.^{1,2} Parker first described the condition in 1845; since then, over new cases have been reported from different parts of the world, but the exact incidence is not known.² It occurs commonly in men in their fourth decade but has been reported in children also, even in neonate.^{3,4}

This is a report of two cases of ISK in patients under 25 years of age, managed by the authors in University of Port Harcourt Teaching Hospital (UPTH), Port Harcourt, Nigeria. This we hope will

increase awareness. Primary anastomosis without any form of bowel preparation can also be done.

2. Presentation of case

2.1. Case 1

The first was a 21-year-old male who presented with a history of sudden generalized abdominal pain of two days duration. He had progressive abdominal distension, nausea and vomiting. He vomited four times prior to presentation. Vomitus consisted of recently ingested meals. He had no urinary symptoms, no fever or jaundice. No previous history of surgery.

Examination revealed a young man in no painful distress but with cold clammy extremities. He was pale, febrile, anicteric but severely dehydrated. His pulse rate was 110 per minute and of small volume. Blood pressure was 90/50 mmHg and respiratory rate 28 cycles per minute. His temperature was 38 °C. His chest was clear clinically. The abdomen was distended, tender and with guarding. Bowel sounds hyperactive. Digital rectal examination revealed an empty rectum. A diagnosis of intestinal obstruction was made.

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Fig. 1. Gangrenous sigmoid (dusky red in appearance).



Fig. 2. Gangrenous ileo-sigmoid knotting.

Packed cell volume done was 43% (14.3 g/dl). Serum electrolyte urea and creatinine results were within normal limits. An erect plain abdominal radiograph showed multiple air fluid levels while the supine showed 3 curved lines of dilated bowel converging to the pelvis.

He was resuscitated with intravenous fluids (normal saline, dextrose infusion). Antibiotics were given (ceftriaxone 1 g, metronidazole 500 mg). Analgesics were also given. Nasogastric tube was passed and Urethral catheterization was done to monitor urine output.

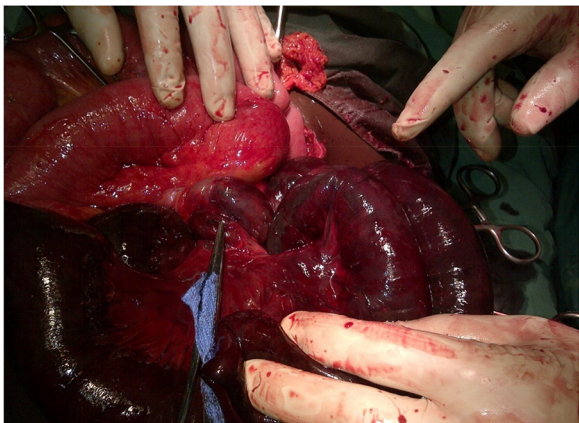


Fig. 3. Point of obstruction showing viable and gangrenous loops of bowel.



Fig. 4. The Patient's distended abdomen.

Within a few hours of admission, he had an exploratory laparotomy.

Findings were as follows: ileo-sigmoid knotting with gangrene of the sigmoid and distal loop of ileum (Figs. 1–3). He had sigmoidectomy with end colostomy (Hartmann's procedure) and right hemicolectomy with ileo-transverse anastomosis.

Three months later the colostomy was closed. His outpatient follow up has been uneventful.

2.2. Case 2

The second case was a 20-year-old male who presented with a history of sudden generalized abdominal pain of three days duration. He had progressive abdominal distension, nausea and vomiting. He vomitted three times in a day prior to presentation. Vomitus consisted of recently ingested meals. He had no urinary symptoms, no fever nor jaundice. No previous surgery.

Examination revealed a young man in no painful or respiratory distress. He was not pale, afebrile, anicteric but mildly dehydrated. His pulse rate was 92 per minute and of good volume. His blood pressure was 110/70 mmHg and respiratory rate 24 cycles per minute. Temperature was 37°C. His chest was clear clinically. Abdomen was distended (Fig. 4), tender and with guarding. His bowel sounds were hyperactive. Digital rectal examination revealed an empty rectum. A diagnosis of intestinal obstruction was made.

Packed cell volume done was 38% (12.4 g/dl). Serum electrolyte urea and creatinine results were within normal limits. An erect plain abdominal radiograph showed multiple air fluid levels while the supine showed 3 curved lines of dilated bowel converging to the pelvis Fig. 5.

He was resuscitated with intravenous fluids (normal saline, dextrose infusion), antibiotics (ceftriaxone 1 gm, metronidazole 500 mg) and analgesics.

Nasogastric tube was passed. Urethral catheterization was done to monitor urine output.

Within a few hours of admission, he had an exploratory laparotomy.

Findings were ileosigmoid knotting with viable loops of bowel Figs. 6–10.

He had careful detorsion, sigmoidectomy with primary anastomosis as his sigmoid colon was redundant Fig. 11.

He did well post-operatively. His outpatient follow up has been uneventful.

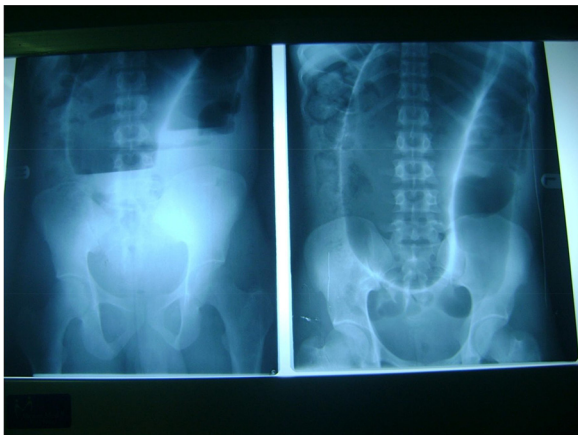
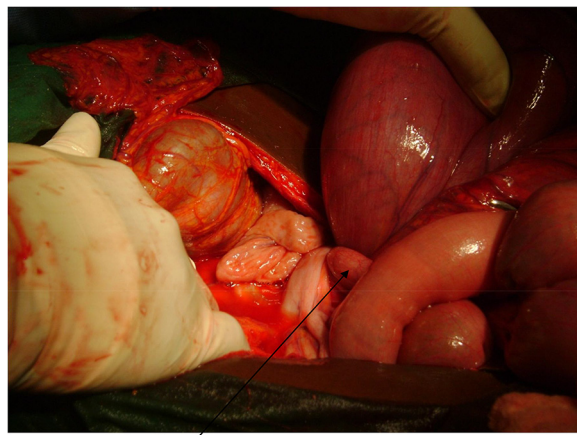


Fig. 5. Plain radiograph of abdomen erect and supine.



Ileum
Fig. 8. Viable ileo-sigmoid knot.

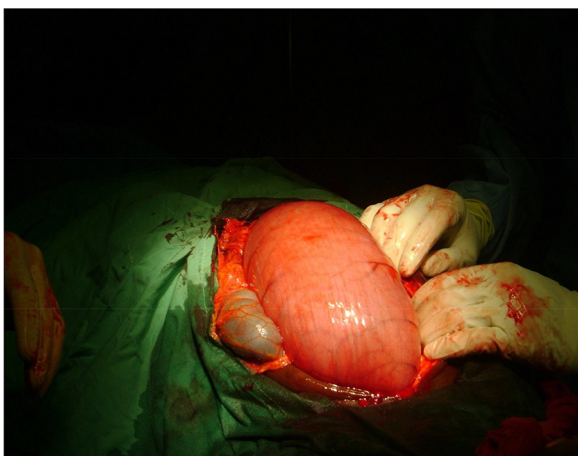


Fig. 6. Viable sigmoid colon.

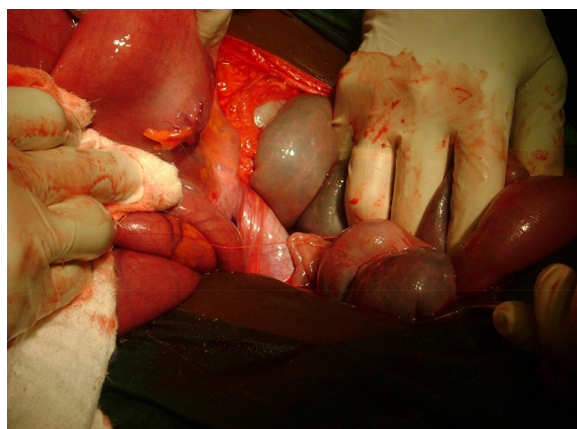


Fig. 9. Viable ileo-sigmoid knot.

3. Discussion

Ileosigmoid knotting is a very rare cause of intestinal obstruction, occurring in men in their fourth decade.³ It is relatively uncommon in females.⁵ It is also referred to as compound volvulus or double volvulus but recently Raveenthiran has suggested that the correct terminology should be compound volvulus.² Jebbin in Mile one Hospital Port Harcourt, Nigeria, reported two cases of

ileo-sigmoid knotting, but this occurred in patient aged 51 and 50 years, respectively.⁶

The aetiology of ISK is not very clear. Some anatomic and dietary factors have mainly been incriminated. These include a long small intestine mesentery with a freely mobile small bowel, a long sigmoid loop on a narrow “V shaped” pedicle, and the consumption of a high bulk diet in the presence of an empty small bowel. Meckel’s diverticulum has been reported to be present in 14–53% of cases⁷ while a relaxed anterior abdominal wall, as commonly seen in post-partum has also been implicated.⁸

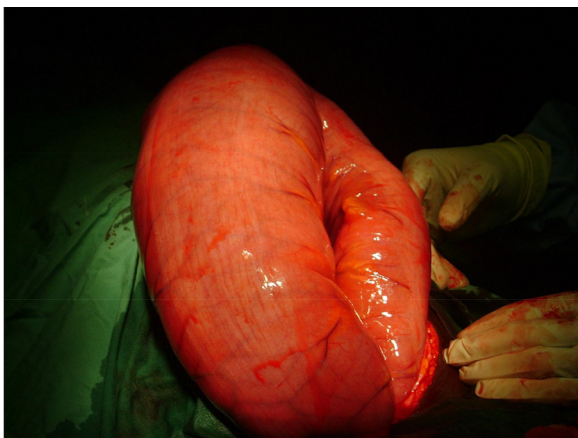


Fig. 7. Viable sigmoid colon.

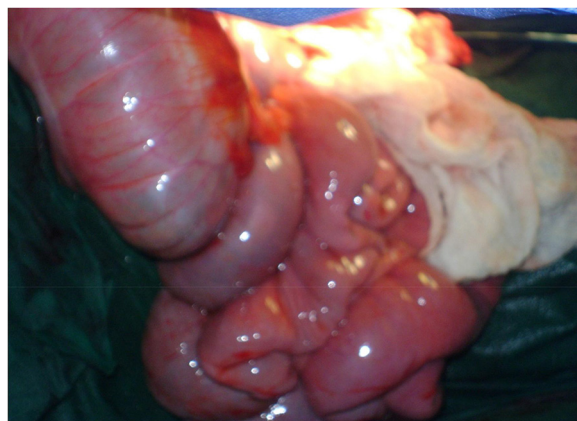


Fig. 10. Viable ileo-sigmoid knot.

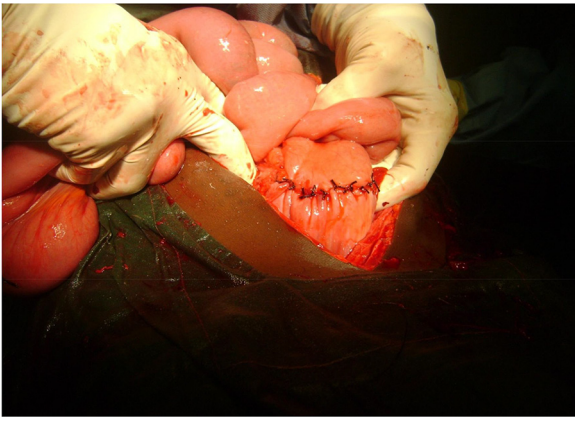


Fig. 11. Primary colo-colic anastomosis following sigmoidectomy.

Shepherd gave a universally accepted description of the pathogenesis of ISK.³ In his description, loops of ileum descend into the left paracolic gutter and wraps around the sigmoid colon, both becoming distended sooner or later. With time, both loops of ileum and sigmoid become strangulated, the latter more frequently so. Gangrene ensues eventually, and may extend to the terminal ileum, sometimes involving the caecum and in very rare occasion, the proximal ascending colon.^{1,3}

The diagnosis of ISK is infrequently made before surgery, this being possible only in about 20% of patients.^{1,8} This is because the features are often nonspecific. Any patient previously in a good state of health and developing a sudden onset of colicky abdominal pain, other features of intestinal obstruction, and in a shock state, all occurring in rapid succession should arouse a suspicion of ISK.^{3,9} The two cases in this report presented in this manner. Suspicion should increase even with younger patients as in index cases.

White blood cell count usually shows leukocytosis due to gangrene¹⁰ and it is thus not specific for ISK. Plain abdominal radiographs may show characteristic double closed loop obstruction, with the sigmoid on the right upper quadrant and the small bowel loops on the left but this is only an occasional finding.^{1,8} More often, the picture is either that of simple sigmoid volvulus or small bowel obstruction but the former is predominant.

Computerized tomography could have diagnostic features of ISK. These have been described in literature. These include the 'whirl sign', created by the twisted intestines and mesentery, and medial deviation of the descending colon with a beak appearance on its medial border.¹¹ This investigation was not done in first case because patient presented in shock and for the second case, the machine was not in a good functional state in Port Harcourt at the time patient presented.

Survival in this rare but serious condition hinges on aggressive resuscitation and early surgical intervention.^{1,12} The diagnosis of intestinal obstruction without a definite pathology is a sufficient indication for a laparotomy. It has been observed that accurate diagnosis is not an end in itself but a means to providing an appropriate treatment.¹³ The surgical option depended on findings at laparotomy. With viable intestinal loops, untying the knot alone is considered sufficient, and recurrence is uncommon.³ Other authors advocate resection of the sigmoid colon in all cases¹⁴ or mesosigmoidoply¹² to avert recurrence. When the bowel loops are gangrenous, most authors recommend the knots should not be unraveled as this can lead to bowel perforation and septic shock.^{1,3} Gangrene of both loops of bowel will demand en bloc resection of the knot and double primary anastomosis of ileal and sigmoid ends.^{3,8} However, if gangrene extends to less than 10 cm from the ileocaecal junction, a right hemicolectomy is

recommended.¹ This was the option adopted in the cases presented in recent times, primary anastomoses after sigmoid resection could be done without any form of bowel preparation as in the second case reported. The patient's condition should be considered. Other options include closure of the distal ileal stump and end-to-end ileocolic anastomosis.^{3,8} The disadvantage of this is that the bypassed portion of the intestine, known as the blind loop, often initiates a cascade of problems known as the blind loop syndrome.¹⁵ Thus it is advised when the patients intra-operative condition is precarious.

There was no consensus in literature as to which bowel should be resected first, ileum or sigmoid? The authors recommend resection of sigmoid en bloc first before the ileum as this may allow recovery of more segments of the later before considering right hemicolectomy.

The prognosis of ISK depends on early diagnosis and prompt surgery.^{2,9} The mortality rate of non-gangrenous ISK ranges from 6.8% to 8%. For gangrenous cases, however, they vary from 20% to 100%.¹⁶ Septic shock is the commonest cause of death.¹⁴ In the case reported by Jebbin, the first patient presented died from ARDS following septic shock. Poor prognostic factors include age over 60 years, duration of symptoms longer than 24 h, and extensive bowel gangrene.¹

4. Conclusion

Ileo-sigmoid knotting is a rare cause of intestinal obstruction. Though this condition is more common in fourth or fifth decade of life, can also occur in the second decade. Its diagnosis often tasks even the clinical acumen of experienced surgeons. This is therefore very often made on table. The prognosis used to be very poor but currently, with early diagnosis, aggressive resuscitation, potent broad spectrum antibiotics and advances in anaesthesia with modern post-operative intensive care, the outcome is improving.

Primary anastomosis of the left colon without any form of bowel preparation can be done especially when patients condition permits.

Previous scientific presentations

First presentation: International College of Surgeons (ICS) Conference Nigerian National Section Ile-Ife 2013.

Second presentation: International College of Surgeons (ICS) Conference Asian Pacific Federation Congress 2013 (APFC held in Thailand).

Conflict of interest

Authors declare no conflict of interest.

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Ethical approval

Written informed consent was obtained from the patients involved.

Author contributions

Case study design and writing was by Author: Patrick Okechukwu Igwe. Contributing authors: N.J. Jebbin, A. Dodiyyi-Manuel, J.M. Adotey.

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References

1. Alver O, Oren D, Tireli M, Kayabasi B, Akdemir D. Ileosigmoid knotting in Turkey. Review of 68 cases. *Dis Colon Rectum* 1993;**36**:1139–47.
2. Raveenthiran V. The ileosigmoid knot: new observations and changing trends. *Dis Colon Rectum* 2001;**44**:1196–200.
3. Shepherd JJ. Ninety-two cases of ileosigmoid knotting in Uganda. *Br J Surg* 1967;**54**:561–6.
4. Chirdan LB, Ameh EA. Sigmoid volvulus and ileosigmoid knotting in children. *Pediatr Surg Int* 2001;**17**:636–7.
5. Scott QJ. Ileosigmoid knot and sigmoid volvulus. *S Afr J Surg* 1973;**11**:29–32.
6. Jebbin NJ. Ileosigmoid knotting: a report of two cases. *Port Harcourt Med J* 2007;**1**:197–200.
7. Kusumoto H, Yoshida M, Takahashi I, Anai H, Maehara Y, Sugimachi K. Complications and diagnosis of Meckel's diverticulum in 776 patients. *Am J Surg* 1992;**164**:382–3.
8. Puthu D, Rajan N, Shenoy GM, Pai SU. The ileosigmoid knot. *Dis Colon Rectum* 1991;**34**:161–6.
9. Odelola MA, Bode CO, Anuma HM. Compound volvulus in the paediatric age group: a report of two cases from Lagos. *Lagos J Surg* 1999;**2**:27–8.
10. Bizer LS, Liebling RW, Delany HM, Gliedman ML. Small bowel obstruction: the role of non-operative treatment in simple intestinal obstruction and predictive criteria for strangulation obstruction. *Surgery* 1981;**89**:407–13.
11. Lee SH, Park YH, Won YS. The ileosigmoid knot: CT findings. *AJR Am J Roentgenol* 2000;**174**:685–7.
12. Akgun Y. Management of ileosigmoid knotting. *Br J Surg* 1997;**84**:672–3.
13. Todd JW. The superior clinical acumen of the old physicians, a myth. *Lancet* 1953;**1**:482–4.
14. Wapnick S. Treatment of intestinal volvulus. *Ann R Coll Surg Engl* 1973;**53**:57–61.
15. Mortensen NJ, Jones O. The small and large intestines. In: Russell RC, Williams NS, Bulstrode CJ, editors. *Bailey and Love's short practice of surgery*. 24th ed. London: Arnold; 2004. p. 1153–85.
16. Hsu ML. Ileosigmoid knot. *J R Coll Surg Edinb* 1979;**24**:28–9.

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