Ileosigmoid Knotting Causing Double Lumen Acute Intestinal Obstruction and Gangrene: Review and A Case Report

Surgery Section

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ABSTRACT

lleosigmoid Knotting is a rare cause of intestinal obstruction. It is also called as compound volvulus or double volvulus. It is caused by the wrapping of the ileum around the sigmoid colon and its mesentery or vice-versa. It is a rapidly progressive condition, leads to acute intestinal obstruction and gangrene in ileum as well as in the sigmoid colon. Early diagnosis and intervention is the key to a better outcome. Due to the rarity and unfamiliarity of this entity, diagnosis is usually made intraoperatively. Surgical removal of the gangrenous segment with either stoma formation or anastomosis is the only hope.

An additional systemic search of the literature was done in PubMed, MEDLINE, ISIS, Embase, and CAS searches with the following free text keywords: ileosigmoid knotting, intestinal knotting, compound volvulus and double volvulus in English literature. Around 64 studies were identified, out of which 38 studies were selected for this article after the removal of duplicates and unrelated articles. These case series and reports were reviewed for aetiopathogenesis, presentation, diagnostic modalities, surgical interventions, and outcome.

Along with this review article, there was a case report of ileosigmoid knotting in a 38-year-old male patient that presented in the surgical Emergency Department; with complaints of generalised pain and distention of abdomen for two days. Also, he had complained of not passing flatus and motion for two days. On examination, patient had generalised tenderness and bowel sounds were absent. X-ray abdomen showed dilated small and large bowel with multiple air-fluid levels. After resuscitation, an emergency exploratory laparotomy was done, and the diagnosis of ileosigmoid knotting with gangrene of both ileum and sigmoid colon was made intraoperatively. After resection of both gangrenous segment, colocolic anastomosis and double barrel ileostomy was performed. The postoperative course was uneventful, and patient was discharged on 7th postoperative day.

Keywords: Gangrenous bowel, Intestinal obstruction, Sigmoid colon

INTRODUCTION

Parker E described the first patient of ileosigmoid knotting in 1845 [1]. The first patient from the Asian subcontinent was reported by Paul M in 1940 [2]. First patient-reported from Africa by Burkitt D in 1952 [3]. The first case from India was reported by Dunkertey GE in 1953 [4]. lleosigmoid knotting, the name first given by Shepherd JJ in 1967 [5]. lleosigmoid knotting is a very rare cause of intestinal obstruction [1]. It occurs due to the wrapping of the ileum or sigmoid colon around the base of the other one, which may cause double-loop intestinal obstruction and/or gangrene of both the loops [6]. It is a well-recognised condition in certain African, Asian, and Middle Eastern nations; but is uncommon in the western world [7]. The patient is usually haemodynamically unstable during presentation due to gangrene of one or both the limb. The patient usually presented with abdominal pain, rapid abdominal distention, and vomiting with rebound tenderness and hyperactive bowel sound. Preoperative diagnosis is difficult because the condition is very rare, and radiographic findings are not able to differentiate from other causes of intestinal obstruction. Usually, diagnosis is made intraoperatively. The most important differential diagnosis is sigmoid volvulus because endoscopic reduction is a contraindication in lleosigmoid knotting. CECT with oral and rectal contrast is helpful in making a preoperative diagnosis but not available in many institutions in emergency settings. Generalised peritonitis with sepsis is the main cause of poor outcome [8]. Immediate surgical intervention with resection of gangrenous part with anastomosis or stoma formation is the only available treatment. This article focuses on the aetiopathogenesis, presentation, diagnostic modalities, surgical interventions, and outcome with the review of articles and case series published to date.

REVIEW OF LITERATURE

Studies and case reports were identified by PubMed, MEDLINE, ISIS, Embase, and CAS searches with the following free text keywords: ileosigmoid knotting, intestinal knotting, compound volvulus and double volvulus in English literature. Other sources included review articles and textbooks. The authors also added a case of ileosigmoid knotting encountered in the surgical emergency.

Data was collected from all available case series and reports from the literature. A total of 38 articles were identified [Table/Fig-1] [1,2,4-39].

Classification

The classification of the disease was made by Alver O et al., in 1993 by taking the ileum and/or sigmoid colon as the active component and considering the direction of the torsion [7].

Type I (55%)- Ileum (active component) wraps itself around the sigmoid colon (passive component)

Type 1a- clockwise direction

Type 1 b- Anticlockwise direction

Type II (25%)- Sigmoid colon (active component) wraps itself around the ileum (passive component)

Type 1a- clockwise direction

Type 1b- Anticlockwise direction

Type III (5%)- The ileocecal segment (active component) wraps itself around the sigmoid colon (passive component)

Type IV Undetermined (15%)- In this, it is impossible to determine the revolved segment

					Age	Sex		Predisposing factors		Alver O et al., Classification (1993)				Investigation		Preoperative diagnosis made		
Serial no.	Year	Author	Total cases	Country	group (mean)	Male	Female	Yes	None	1	Ш	Ш	IV	X- ray	CT scan	Yes	No	Outcome
1	1845	Parker E [1]	1	UK	53	1	0	0	1	1	0	0	0	NA	NA	NA	NA	NA
2	1932	Kallio KE [9]	1	Finland	48	1	0	0	1	1	0	0	0	NA	NA	NA	NA	Alive
3	1940	Paul M [2] Dunkerley	1	Ceylon	NA	0	1	1	0	1	0	0	0	NA	NA	NA	NA	Alive
4	1953	GE [4]	1	India	61	1	0	0	1	NA	NA	NA	NA	NA	NA	NA	NA	Died
5	1967	Shepherd JJ [5] Roy SP et al.,	92	Uganda	42	78	14	12	80	60	19	1	12	NA	NA	NA	NA	20-died
6	1973	[11]	9	India	39	7	2	2	7	NA	NA	NA	NA	8	0	0	9	1-died
7	1981	Kakar A and Bhatnagar BN [12]	11	India	38	6	5	2	9	NA	NA	NA	NA	11	0	0	11	NA
8	1984	Watson RG [13]	7	South Africa	NA	NA	NA	1	6	NA	NA	NA	NA	5	0	0	7	2-died
9	1986	Johnson CD [14]	1	UK	NA	NA	NA	0	1	1	0	0	0	1	0	0	1	Died
10	1991	Puthu D et al., [8]	7	India	40	4	3	1	6	5	1	0	1	7	1	1	6	4-died
11	1992	Guessan HA et al., [10]	16	Abidjan	42	10	6	1	11	NA	NA	NA	NA	NA	NA	NA	NA	7-died
12	1993	Gibney EJ and Mock CN [15]	15	Ghana	NA	11	4	1	14	NA	NA	NA	NA	14	2	0	15	2-died
13	1993	Alver O et al., [7]	68	Turkey	49	57	11	10	58	45	12	3	8	62	3	2	66	14-died
14	1997	Akgun Y [16]	16	Turkey	45	11	5	2	14	8	3	1	4	13	0	1	15	3-died
15	1998	Kedir M et al., [17]	1	Ethiopia	NA	NA	NA	0	1	NA	NA	NA	NA	NA	NA	NA	NA	NA
16	2001	Raveenthiran V [18]	7	India	43	6	1	1	6	5	0	0	2	6	0	NA	NA	3-died
17	2002	Nasir GAA and Mohammed MM [19]	7	Iraq	NA	5	2	0	7	4	1	0	2	7	0	0	7	1-died
18	2004	Hashimoto T et al., [20]	2	Japan	32	2	0	0	2	2	0	0	0	NA	NA	0	2	1-died
19	2004	Atamanalp SS et al., [6]	63	Turkey	47	47	16	6	57	49	5	1	8	57	2	1	62	13-died
20	2007	Jebbin NJ [21]	2	Nigeria	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	All alive
21	2008	Bawa D et al., [22]	1	Nigeria	26	1	0	0	1	NA	NA	NA	NA	1	0	NA	NA	NA
22	2009	Machado NO [23]	1	Muscat	52	1	0	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	Alive
23	2009	Zahid FE et al., [24]	1	Morocco	42	1	0	NA	NA	NA	NA	NA	NA	1	0	NA	NA	Died
24	2009	Alvi AR et al., [25]	2	Karachi	44	2	0	1	1	2	0	0	0	NA	NA	0	2	1-died
25	2009	Atamanalp SS et al., [26]	71	Turkey	37	50	21	20	51	52	8	1	10	66	2	1	70	20-died
26	2009	Okello TR et al., [27]	44	Uganda	39	36	8	8	36	34	4	2	4	35	0	0	44	13-died
27	2011	Ahmadi nazad M et al., [28]	1	Iran	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	Alive
28	2011	Baheti AD et al., [29]	1	India	51	1	0	NA	NA	NA	NA	NA	NA	1	0	0	1	Alive
29	2011	Atamanalp SS [30]	32	Turkey	41	21	11	11	21	21	6	1	4	30	1	1	31	7-died
30	2014	Banerjee C [31]	9	India	47	8	1	2	7	NA	NA	NA	NA	9	0	NA	NA	2-died
31	2012	Uday SK et al., [32]	1	India	32	1	0	NA	NA	1	0	0	0	1	0	0	1	Alive
32	2013	Kumar S et al., [33]	1	India	46	1	0	NA	NA	1	0	0	0	1	0	0	1	Alive
33	2014	Igwe PO [34]	2	Nigeria	21	2	0	0	2	2	0	0	0	2	0	0	2	Alive
34	2014	Andromanakos N et al., [35]	1	Greece	NA	NA	NA	0	1	NA	NA	NA	NA	NA	NA	NA	NA	Died
35	2014	Shimizu R et al., [36]	1	Japan	NA	NA	NA	0	1	NA	NA	NA	NA	1	0	NA	NA	Alive

36	2014	Bhambare M et al., [37]	1	India	NA	NA	NA	1	0	NA	NA	NA	NA	1	0	0	1	Alive
37	2014	Yazough I et al., [38]	1	Africa	61	0	1	NA	NA	1	0	0	0	1	0	0	1	Alive
38	2016	Islam S et al., [39]	2	Pakistan	NA	NA	NA	1	1	NA	1-died							
[Table/Fig-1]: Different case reports and series of ileosigmoid knotting described in literature till date [1,2,4-39].																		

A new classification was developed by Atamanalp SS et al. [26] in 2008 for ileosigmoid knotting, using mortality criteria, such as age over 60 years, and the presence of accompanying disease or shock. In this proposed classification, the patients were classified as follows:

Class 1: Patients with no risk factor;

Class 2: Patient with no shock or gangrene but with other risk factors, such as age over 60 years, or existence of an associated disease;

Class 3: Patient with shock;

Class 4: Patient with single segment bowel gangrene (ileum or sigmoid colon);

Class 5: Shock and single-segment bowel gangrene;

Class 6: Patient with gangrene in both segments.

Depending on the presence of other risk factors, Class 2, 3, and 4 were further divided into sub-groups of a and b. They studied 71 patients with ileosigmoid knotting that were treated surgically in the Department of General Surgery, School of Medicine, Atatürk University in 42 years period between June, 1966 and July, 2008 [26].

Epidemiology

Ileosigmoid knotting has a higher incidence in East Africa, but it is reported from other parts of the world like Asian, Middle Eastern, Eastern, Northern European, South American countries, and also from Turkey [12-20]. It more commonly affects males who are in the fourth decade of life [5]. Geographical location, racial predilection, habit and diet make the condition common among Africans [10].

Aetiology

Three factors are responsible for the ileosigmoid knotting [7,8,12]

- 1- A long small bowel mesentery with freely mobile small bowel
- 2- Ingestion of a high bulk of the diet in the presence of an empty small bowel
- 3- A long sigmoid colon on a narrow pedicle

Secondary causative factors which are also described in the literature are: late pregnancy, Meckel diverticulitis with a band, transmesenteric herniation, ileo-cecal intussusceptions, and floating cecum [7,8]. The consumption of a high bulk diet in the presence of an empty small bowel can be a predisposing factor. Therefore, the incidence is high among Muslims who eat a single daily meal during the Ramadan fast [7].

Pathogenesis

The pathogenesis of ileosigmoid knotting is not clear. Suggested theory is, a semi-liquid bulky meal passing from proximal jejunum to the rest of the bowel, increases the mobility of the rest of the bowel. This causes the heavier segments of the proximal jejunum to shift into the left lower quadrant. Remaining empty loops of ileum and distal jejunum twist in a clockwise rotation around the base of a narrow sigmoid colon. Progressive peristalsis forms an ileosigmoid knot with two closed-loop obstruction in the small bowel and sigmoid colon [7].

In ileosigmoid knotting, there is an obstruction in both ileum and sigmoid colon. Progressively, it causes strangulation, and thrombosis of the vessels leading to ischemia and gangrene in the ileum and sigmoid colon. The gangrene may extend to the proximal part of the ileum, the cecum, and rarely the distal part of the jejunum and ascending colon [7,8]. Obstruction or gangrene causes the release of endotoxin and bacterial translocation in the peritoneal cavity resulting in septicaemia and shock [6-8].

Clinical Features

The patient is usually haemodynamically unstable during presentation due to gangrene of one or both the limbs. The patient usually presented with abdominal pain, rapid abdominal distention, and vomiting. Other complaints include nausea, diarrhoea, anorexia, rectal bleeding, and hematemesis. On examination, the main signs are asymmetrical abdominal distention and generalised tenderness. Additional findings include hyperactive bowel sounds, tympanic sound all over the abdomen, ballooning of rectum, and visible peristalsis [7,8]. It can rapidly progress to gangrene of the ileum and/or sigmoid colon [8]. If not treated immediately, patient may go in generalised peritonitis, sepsis, and dehydration leading to death. In this review of published articles the following observations were made: abdominal pain and tenderness (100%, n-38) [1-2,4-39], abdominal distension (95%, n-37) [1-2,5-39], nausea and vomiting (87%, n-36) [1-2,4-37], rebound tenderness (70%, n-34) were identified [1-2,5-36].

Diagnosis

Preoperative diagnosis is difficult because the patient usually presents with non-specific symptoms in an emergency that mimic the features of small and/or large bowel obstruction. It should be suspected in patients with clinical features suggestive of small bowel obstruction, while the radiographic findings are that of colonic obstruction [18]. Laboratory reports usually show a drop in haemoglobin, leukocytosis, raised Blood Urea Nitrogen (BUN), and electrolyte imbalance suggestive of peritonitis [6]. Plain abdominal X-ray radiographs demonstrate a dilated sigmoid colon and multiple small intestinal air-fluid levels. The X-ray may show free gas under diaphragm due to the perforation of any segment [6,7].

Clinically or on plain X-ray, it is often mistaken for sigmoid volvulus. Sigmoidoscope or a flatus tube may not deflate the distended colon or help to release the ileosigmoid knotting [8,12]. CECT with oral and rectal contrast can help in preoperative diagnosis, but due to the rarity of this condition, even CECT may fail to reach the correct diagnosis [40]. Also, CECT is not available in many institutions in emergency settings. The findings in a CECT scan suggestive of ileosigmoid knotting include the whirl sign. It is created by twisted intestine and sigmoid mesocolon in ileosigmoid knot, medial deviation of the cecum, and descending colon [40,41]. Signs of bowel ischaemia or gangrene caused by strangulation can be seen in a CT scan [42,43]. A flexible sigmoidoscopy can demonstrate a spiral sphincter-like twisting of the mucosa in the sigmoid colon, but it does not give any information about the small bowel [42].

Based on a review of the literature, the fulfillment of these three conditions may reach to correct diagnosis of ileosigmoid knotting [16,18,22,41].

- Clinical symptoms of small bowel obstruction like predominant vomiting.
- 2. X-ray or CECT evidence of predominately large bowel obstruction involving the sigmoid colon
- 3. Inability to insert a sigmoidoscope or flatus tube to relieve symptoms.

If all three features are present, one should raise the suspicion of ileosigmoid knotting.

Treatment

The patient is usually haemodynamically unstable on presentation. So, priority should be given for nasogastric decompression, aggressive resuscitation with fluid and electrolytes with the help of central venous pressure monitoring, or wide bore cannula [6,7]. Correction of acid-base imbalance is very important in these patients as vomiting can be present with hyponatremia and hypokalemia [6,7]. Exploratory laparotomy is the only available treatment and should not be delayed after haemodynamic stabilisation [7]. Broad-spectrum antibiotic therapy is given early and continued after the surgery. The usual antibiotic combination includes cephalosporins/imipenem group, aminoglycosides, and metronidazole [16].

Surgical procedures depend on the viability of single or both loops. Knot may be untwisted if both loops are viable. It can be done by sigmoid enterotomy or needle puncture, if not able to do without it, when the ileum and the sigmoid colon both are gangrenous, the chance of spillage of toxic bowel contents is there during the untying of knot [16]. Intestinal clamps should be applied before untwisting or resection of the knot followed by resection of the gangrenous loop to minimise contamination. The primary anastomosis can be done if both ends of the small or large bowel are viable and bleed well [16]. Hartmann operation or a covering colostomy was recommended previously to avoid the risk of faecal leak from the colonic anastomosis, but recent literature said that primary colonic anastomosis might be undertaken safely when history is short, and the remaining bowel is cleaned and well-vascularised [16].

Primary anastomosis in the gangrenous sigmoid colon can be safely carried out without significant complications [44,45]. Second look laparotomy and anastomosis can be carried out if both ends of the bowel are healthy, had no gross contamination, with a rich blood supply, and if a tension-free anastomosis could be achieved [11,44,45]. In patients with blood pressure <90/60 mm Hg and a pulse >120 beats/min, either preoperatively, or after the induction, despite fluid resuscitation, laparotomy should be considered. This involves en-block resection of the involved segment closure of the bowel edges peritoneal lavage and temporary abdominal closure [46]. As a precaution, a stoma should be used to retain the viability of the stump [44]. Intraoperative colonic irrigation and peritoneal lavage is mandatory even with minimal faecal contamination [18].

Prognostic Factors

The reported mortality in ileosigmoid knotting varies from 2% to 55% (mean 36%) in different studies [16,18,21,22,44,45]. Various prognostic factors are described, out of which the general condition of the patient, presence of shock, presence of gangrene, and total duration of disease are important [47]. The mortality rate is high in gangrenous cases [48]. The morbidity rate is also high [48]. The most common cause of death is shock [42]. A second look surgery or surgery with stoma formation has a high survival rate but more morbidity [16]. A review of the literature revealed that there has been a constant decline

in mortality in the last 20 years due to better knowledge of the condition, broad-spectrum antibiotic availability, ICU care, and newer surgical skills [22].

CASE REPORT

A 38-year-old Muslim male patient, was brought in Surgery Emergency Department with a history of generalised abdominal pain, abdominal distention, bilious vomiting, and non-flatus of flatus and stools for two days. The patient also had a history of pulmonary tuberculosis six years back, for which he received Antitubercular Therapy (ATT) for six months.

On examination, his pulse rate was 120/min, BP-80/50 mmHg. Bilateral wheeze were present in the chest. The abdomen was tense and distended with generalised tenderness and guarding. Bowel sounds were absent. Rebound tenderness was present, and on per rectal examination, soft faecal matter was present.

Lab investigation shows hypokalemia and hyponatremia due to excessive vomiting. X-ray erect abdomen revealed multiple airfluid levels with dilated large bowel suggestive of both small and large bowel obstruction [Table/Fig-2]. There was no gas under the diaphragm in the chest X-ray. Ultrasonography showed multiple dilated bowel loops with minimal interbowel fluid. CECT was not available in an emergency setting.



[Table/Fig-2]: X-ray shows dilated small and large bowel with an air-fluid level.

After aggressive resuscitation, an urgent exploratory laparotomy was done. Intraoperative findings are: sigmoid colon mesentery was wrapped around ileum (type-2 classification), making two complete turns with gangrene of both sigmoid colon and ileum [Table/Fig-3,4]. Clamps applied, and both gangrenous sigmoid colon and ileum were resected. Double barrel ileostomy with colocolic anastomosis was done because the viability of both ends of ileum was doubtful, and the patient was haemodynamically unstable. The patient remained on ventilator support for one day. However, he improved gradually. Stoma became functional on the second postoperative day, and then he was orally allowed. The patient was discharged after one week with a plan of ileostomy closure after six weeks.





CONCLUSION(S)

lleosigmoid knotting rarely causes intestinal obstruction. Early diagnosis and intervention are necessary for a better outcome. Clinical signs of small bowel obstruction, radiological signs of large bowel obstruction, and failure of hydrostatic reduction may raise the suspicion of lleosigmoid knotting. But in most of the cases, diagnosis is made intraoperatively. This condition should be differentiated from sigmoid volvulus because it is not relieved by hydrostatic reduction or sigmoidoscopy. Aggressive management with fluid resuscitation, preoperative antibiotics, early laparotomy, optimum surgical technique, and better perioperative care has increased the chance of survival of these patients. In the past, it has a bad prognosis, but now these figures are improving due to the above-mentioned factors.

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AUTHOR DECLARATION:

- Financial or Other Competing Interests: None
- Was Ethics Committee Approval obtained for this study? NA
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

PLAGIARISM CHECKING METHODS: [Jain H et al.]

- Plagiarism X-checker: May 20, 2020
- Manual Googling: Jul 15, 2020
- iThenticate Software: Sep 30, 2020 (21%)

ETYMOLOGY: Author Origin

Date of Submission: May 20, 2020 Date of Peer Review: Jun 11, 2020 Date of Acceptance: Jul 16, 2020 Date of Publishing: Oct 01, 2020