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Case Report

CASE REPORT

Ileosigmoid knotting: a rare case report with review of literature†

Mukesh Sangwan[1], Vijayata Sangwan[2,*], M.K. Garg[1], Jagdish Mutreja[3], Deepak Singla[1], and Dinesh Gautam[1]

[1]Department of General Surgery, B.P.S. Govt. Medical College for Women, Sonepat, Haryana, India, [2]Department of Obstetrics and Gynaecology, B.P.S. Govt. Medical College for Women, Sonepat, Haryana, India, and [3]E.S.I. Hospital, New Delhi, India

*Correspondence address. Department of Obstetrics and Gynaecology, B.P.S. Govt. Medical College for Women, Khanpur Kalan, Sonepat, Haryana 131305, India. Tel: 01263-283402; Fax: 01263-283064; E-mail: vsangwan03@gmail.com

Abstract

Ileosigmoid knotting (ISK) is a rare cause of intestinal obstruction in which loops of ileum and sigmoid colon wrap around each other. It is very uncommon in western world when compared with the African and Asian region. It is rapidly a progressive, fatal disease. Early diagnosis and intervention is the key of better outcome. We are reporting a case of 51-year-old male who presented with shock within 24 h of onset of symptoms. Exploratory laparotomy revealed ISK causing gangrene of ileum and sigmoid colon. In view of haemodynamic unstability, end ileostomy was done after excising gangrenous segments. The patient expired after 2 weeks due to complications of short bowl syndrome. We are also tabulating all cases of ISK reported in the literature till date.

INTRODUCTION

Ileosigmoid knotting (ISK) is a rare cause of closed-loop intestinal obstruction, which rapidly progresses to gangrene of involved gut segments [1]. ISK is a very unusual entity in western world, but is relatively common in Asian, Middle Eastern and African nations [2]. It is associated with 0.5–1.7% of intestinal obstruction [3]. Although the reported mortality rate in ISK varies from 0 to 48% (mean 35.5%), but ISK with gangrene has a mortality rate [1, 3] of 20–100%.

Parker described the first case of ISK in 1845 [1, 4]. Kallio reported the second case in [5] 1932. Paul described the first case of ISK from Asian subcontinent in 1940. More than 330 cases in world and only 22 cases from India have been described so far in the literature as summarized in Table I. It is still a diagnostic dilemma for surgeons all over the world and only 0–28% cases could be diagnosed preoperatively. X-ray abdomen can reveal large gas-filled loops of small and large bowels in the right mid and lower abdomen. Ultrasonography reveals dilated fluid-filled gut loops and free fluid in the pelvis. Computed tomography (CT) is the best diagnostic modality in clinching the diagnosis.

CASE REPORT

A 51-year-old male was brought in our casuality in shock with history of abdomen pain and non-passage of flatus and stools for 24 h. His blood pressure was 78/56 mmHg and pulse rate was 112 min^{-1}. Abdomen was distended and tender. He was resuscitated, and X-ray of abdomen revealed multiple dilated gut

† This case report is regarding diagnostic dilemma and management of ileosigmoid knotting, which is a very rapidly progressive fatal entity if not diagnosed timely.

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Table 1: Review of ISK cases

S. no.	Author	Year	No. of cases	Country
1	Parker	1845	1	—
2	Kallio	1932	1	—
3	Paul	1940	1	India
4	Guessan	1962	16	Abidjan
5	Shepherd	1967	92	Uganda
6	Roy	1973	9	India
7	Watson	1984	7	South Africa
8	Johnson	1986	1	United kingdom
9	Puthu	1991	7	India
10	Gibney	1993	15	Ghana
11	Alver	1993	68	Turkey
12	Akgun	1997	16	Turkey
13	Mohammed	1998	1	Ethiopia
14	Ghassan	2002	7	Iraq
15	Atamanalp	2004	63	Turkey
16	Atamanalp	2007	9	Turkey
17	Jebbin	2007	2	Nigeria
18	Bawa	2008	1	Nigeria
19	Ugwu	2008	1	Nigeria
20	Machado	2009	1	Muscat
21	Zahid	2009	1	Morocco
22	Alvi	2009	2	—
23	Atamanalp	2009	71	Turkey
24	Islam	2009	2	—
25	Okello	2009	44	Uganda
26	Ahmadinazad	2010	1	—
27	Baheti	2011	1	India
28	Atamanalp	2011	32	Turkey
29	Babu	2011	1	India
30	Uday	2012	1	India
31	Kumar	2013	1	India
32	Andromanakos	2014	1	Greece
33	Shimizu	2014	1	Japan
34	Bhambari	2014	1	India
35	Igwe	2014	1	—
36	Darnkeith	2014	2	—
37	Yazough	2014	1	Africa

loops as depicted in Fig. 1. Per rectal examination revealed soft faecal matter. Ultrasonography showed multiple dilated gut loops with minimal intergut fluid. CT was not available. Urgent exploratory laparotomy was done. Ileum was wrapped around sigmoid colon making two complete turns with gangrene of sigmoid colon and ileum as depicted in Fig. 2. Clamps applied on large gut and sigmoid colon removed followed by ileum, and end jejunostomy was done in view of haemodynamic unstability of the patient along with descending sigmoid colon anastomosis. The patient expired after 2 weeks despite exhaustive efforts due to complications of short bowl syndrome.

DISCUSSION

The ISK is rare but life-threatening type of closed-loop intestinal obstruction. The exact mechanism of ISK is still speculative. A long small bowel mesentery, long sigmoid mesocolon on a narrow pedicle and ingestion of high bulk diet after fasting are predisposing factors [1, 6]. The ileal loops can twist around sigmoid colon in clockwise (60.9–63.2%) or anticlockwise direction (36.8–39.1%). The knot is 360° in 52.9%, two 360° turns in 19.1% and three 360° turns in 5.9% cases. ISK is seen predominantly in males (80.2%), with a mean age of 40 years (4–90 years).

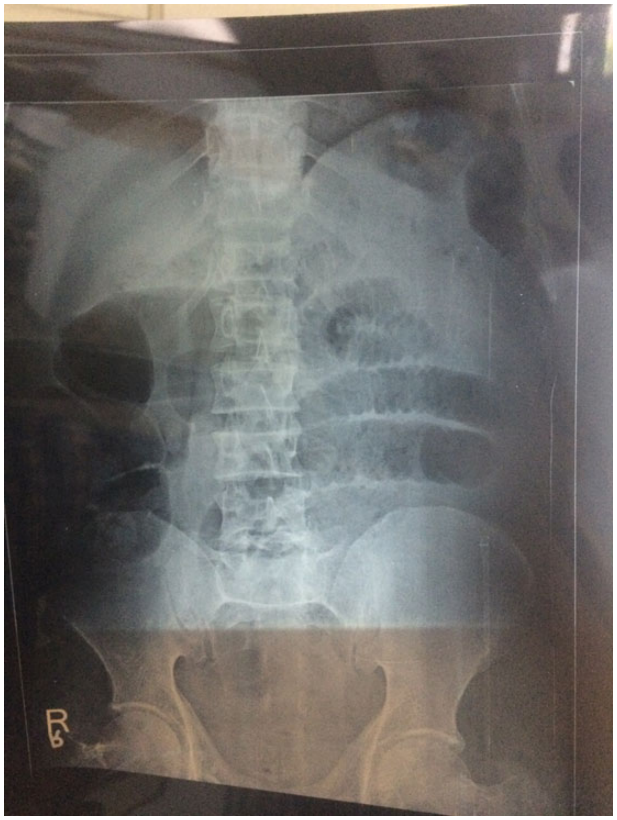

Figure 1: X-ray abdomen showing dilated small and large gut loops.

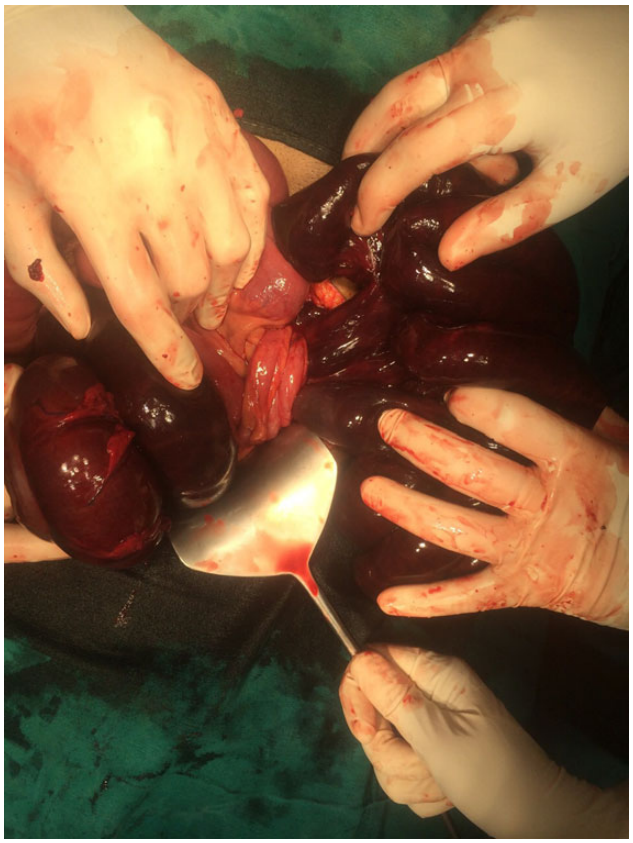

Figure 2: Intraoperative picture showing ileal knotting around sigmoid colon resulting in gangrene of both.

Table 2: Classification of ISK [1, 2, 7]

S. no.	Type of ISK	Percentage	Active component	Passive component	Type of rotation
1	Type 1A	53.9–57.5	Ileum	Sigmoid colon	Clockwise
	Type 1B				Anticlockwise
2	Type 2A	18.9–20.65	Sigmoid colon	Ileum	Clockwise
	Type 2B				Anticlockwise
3	Type 3	1.5	Ileocaecal segment	Sigmoid colon	—
4	Type 4	—	Undetermined	—	—

Table 3: New classification for ISK [8]

C1	C2a	C2b	C3a	C3b	C4a	C4b	C5	C6
A0 D0	One of A, D1	Two of A, D1	At most 1 of A, D1	Two of A, D1	At most 1 of A, D1	Two of A, D1	—	—
S0	S0	S0	S1	S1	S0	S0	S1	—
G0	G0	G0	G0	G0	G1	G1	G1	G2

C: class; A (age): A0: under 60 years; A1: 60 years and older; D (associated disease): D0: absent; D1: present; S (shock): S0: absent; S1: present; G (bowel gangrene): G0: absent; G1: present in the ileum or sigmoid colon; G2: in both segments.

ISK has been classified into four types as described in Table 2. In the present case, we have Type IA knot (most common variety) with two 360° turns of ileum over sigmoid colon. Atamanalp *et al*. in 2008 described a new classification of ISK based on age, shock, associated chronic illness and shock as summarized in Table 3. ISK is a known entity for its rapid progression to gangrene. Pain abdomen (100%), abdomen distension (94–100%), nausea and vomiting (87–100%) and shock (0–60%) are usually present at admission [1, 2, 9]. X-ray abdomen reveals a large gas-filled loop of sigmoid colon in the right mid and lower abdomen. Ultrasonography shows dilated gas-filled intestinal loops with free fluid. CT of abdomen can help to clinch the diagnosis in preoperative stage [1, 2]. It can markedly reveal a dilated loop of sigmoid colon with loss of haustration and non-enhancing thinned out wall. The characteristic whirl sign (twisted mesentery and bowel) is another feature. Convergence of superior mesenteric vein towards knot and medial pointing of caecum are other suggestive findings. Despite availability of various modern diagnostic tools in the present era, only 0–28% cases could be diagnosed preoperatively [3, 10].

Exploratory laparotomy is the definite key to diagnose majority of these cases of diagnostic dilemma. Gut gangrene is present in 73.5–79.4% cases of ISK. Both ileum and sigmoid were gangrenous in 52.9–60.3% of the cases. The incidence rate of gangrene was paradoxically high (90.9%) in cases who presented within 24 h of onset of their symptoms than those presenting after 24 h of their symptoms [1] (57%) as happened in the present case also. Excision of gangrenous segment with end-to-end anastomosis of healthy bowel is the most acceptable surgical procedure in the literature [1, 2]. Hartmann's sigmoidectomy and end colostomy are other accepted options.

ISK is a rare entity with grave prognosis. A high index of suspicion is required for all these cases. In view of diagnostic dilemma and rapidly fatal course of disease, urgent exploratory laparotomy is the best answer for better outcome. While selecting the type of surgery, general condition of patient must be kept in centre.

CONFLICT OF INTEREST STATEMENT

M.S. as the main and corresponding author certifies all authors that this paper is original and had not been sent to any other journal for publication. There is no conflict of interest among the authors. M.S. wish to state that as in this case report, no research work is conducted so institutional ethical committee involvement was not done. Informed consent of the patient was taken, and he was acknowledged orally regarding the process and ensured that his identity will not be revealed anywhere.

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