Adult Intussusception: An Uncommon Condition and Challenging Management

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Keywords

Intussusception · Adult · Diagnosis · Management · Surgery

Abstract

Introduction: Intussusception is a rare condition in adults. A pathological lesion is usually found with a significant percentage of malignancy. The optimal treatment is still not universally clear. **Methods:** This is a retrospective review of adult patients with a diagnosis of intestinal intussusception and surgically treated at our institution from January 2009 to December 2018. Clinical, operative, and histological details were collected and analyzed. **Results:** A total of 26 cases, 16 males and 10 females, were diagnosed with surgically proven intussusception during the 10-year period. The mean age was 45 years (range 21–70). Using ultrasound and/or computed tomography as imaging study, the preoperative diagnosis was made in 21/26 (81%) patients. Five intussusceptions were discovered only upon exploratory laparotomy for intestinal obstruction. There were 19 (73%) cases of enteric and 7 (27%) cases of colonic intussusceptions. All patients underwent surgical exploration. Intestinal resection with immediate anastomosis was the technique of choice for most patients. A single patient underwent stoma for peritonitis secondary to intestinal perforation. An organic cause has been systematically revealed, and no idiopathic intussusception was detected. Etiology was malignant in 9 (35%) cases. **Conclusion:** Adult intussusception should be considered in any patient with subacute abdominal pain. Considering the

high rate of malignancy, intestinal resection without attempting reduction is highly recommended for colonic intussusceptions. However, a more selective approach can be adopted for enteric intussusceptions.

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Introduction

Intussusception, first described by Paul Barbette in 1674, is defined as the invagination of an intestinal segment (intussusceptum) into the lumen of another adjacent segment (intussusciens) [1, 2]. It can lead to intestinal obstruction and exposes the patient to the risk of parietal ischemia, perforation, and peritonitis. Intussusception in adults is entirely different from the pediatric form, especially regarding clinical, pathological, and therapeutic aspects. Intussusception in children is a common disease, usually idiopathic without an identified lead point. Clinical presentation is often acute and easily recognized by the classic triad of abdominal pain, rectal bleeding, and a palpable abdominal mass. Most cases can be successfully treated by hydrostatic or pneumatic reduction without surgical intervention. In contrast, intussusception in adults is a rare disorder that represents only 1% of intestinal obstructions and nearly 5% of all intussusception cases [3–5]. Unfortunately, clinical signs are often nonspecific and make diagnosis difficult. Nevertheless, the widespread use of computed tomography (CT) for abdominal complaints has significant-

Fig. 1. Ileo-colic intussusception due to cecum adenocarcinoma in a 30-year-old woman. Axial contrast-enhanced CT scan showing a typical round target-shaped mass with bowel wall thickening.

Fig. 2. Enteric intussusception due to fibrous polyp in a 58-year-old man. Contrast-enhanced CT scan shows a sausage-shaped mass with edematous bowel wall in the ileal loop. Mesenteric vessels and fat tissues are seen in the center of the mass.

Table 1. Symptoms of patients with intussusception

Symptoms	n/N (%)
Abdominal pain	24/26 (92)
Nausea/vomiting	22/26 (85)
Abdominal distension	10/26 (38)
Constipation	9/26 (35)
Palpable abdominal mass	7/26 (27)
Diarrhea	6/26 (23)
Weight loss	5/26 (19)
Anorexia	3/26 (12)
Rectal bleeding	3/26 (12)
Fever	2/26 (8)

ly increased the detection of adult intussusception, especially transient and asymptomatic forms. The typical bowel-within-bowel appearance often containing mesenteric fat and blood vessels is pathognomonic. Most adult intussusceptions are due to pathological lead point with approximately 60% of malignancy [6–8]. Usually, surgical exploration is needed to verify the diagnosis and exclude an underlying malignancy. However, there is no universal consensus about indications of intraoperative reduction and the extent of intestinal resection.

This study reports our experience and discusses clinical features, diagnosis, and optimal management of this uncommon entity.

Materials and Methods

A retrospective study was conducted over a period of 10 years. This study included all adult patients (aged 18 years or older) treated for intestinal intussusception at Mohammed V Military Hospital of Rabat, Morocco, from January 2009 to December 2018. Sur-

gical data were from patients' medical records. Clinical presentation, investigation findings, operative details, and histological reports were collected and analyzed.

Patients were divided into 2 groups according to the lead point location: enteric intussusceptions and colonic intussusceptions. The enteric form is confined to the small bowel, including jejuno-jejunal, jejuno-ileal, and ileo-ileal intussusceptions. The colonic form includes colocolonic and colorectal intussusceptions. Ileocolic intussusception is considered as enteric if the lead point is ileal (proximal to the ileo-cecal valve) and as colonic if the lead point is colonic (distal to the ileo-cecal valve). Rectal prolapse was excluded from this classification. Anterograde intussusception was defined by telescoping of an intestinal segment into the lumen of another proximal segment. Inversely, retrograde intussusception corresponded to telescoping of an intestinal segment into the lumen of another distal segment. Acute symptoms were defined as symptoms that typically lasted <4 days, as subacute when lasting from 4 to 14 days, and as chronic when lasting >14 days.

Intussusception was preoperatively diagnosed by abdominal ultrasound and/or CT scan. All patients underwent surgical treatment using an open or laparoscopic approach, and the timing of surgery depended on the urgency of the patient's clinical presentation.

Results

Age and Gender Data

A total of 26 patients, 16 males and 10 females, were diagnosed with adult intussusception during the 10-year period. The mean age was 45 years, with a range of 21–70 years. The male-to-female ratio was 1.6:1.

Clinical Presentation

Out of the 26 patients, 8 (31%) were admitted to the emergency department with clinical signs of acute intestinal obstruction, and 1 of them had peritonitis. The other 18 (69%) had subacute to chronic symptoms suggestive of subacute obstruction, such as episodic abdominal pain and nausea/vomiting. A palpable mass was found in 7 pa-

Fig. 3. Ileo-ileal intussusception due to adenomyoma in a 25-year-old man. Contrast-enhanced CT scan showing the pathognomonic “target sign” on the sagittal view (**A**) and the typical “sausage-shaped sign” on the frontal view (**B**).

tients at abdominal examination, but none had the classic pediatric triad. The time from symptoms onset to diagnostic confirmation varied from 1 day to 2 years. Details of symptoms and clinical signs are listed in Table 1.

Diagnostic Modalities

Plain abdominal radiography, performed only in patients with acute clinical presentation, showed hydro-air levels typical of intestinal obstruction. Abdominal ultrasound was done in 17 patients, which revealed classical images (target sign or pseudokidney sign) highly suggestive of intestinal invagination in 9 of them (53% accuracy). Abdominal CT scan, undertaken in 23 patients, confirmed intussusception in 21 patients (91% accuracy; Fig. 1–3). However, a pathological lead point was found in only 10 patients out of the 21 (48%). Total colonoscopy, performed in 3 patients, revealed intussusception secondary to cecum adenocarcinoma in 1 patient (Table 2). Moreover, 5 cases were discovered only upon exploratory laparotomy for intestinal obstruction. Overall, preoperative diagnosis of intussusception was made in 21/26 (81%) patients using the above-mentioned modalities.

Treatment and Postoperative Course

All patients underwent surgical intervention. Eight of them (31%) were admitted with acute intestinal obstruction and required emergency surgery. The other nonurgent patients ($n = 18$, 69%) were operated on the basis of elective surgery. Open laparotomy was usually used as a surgical approach, whereas laparoscopic surgery was suc-

Table 2. Preoperative diagnostic studies

Study	Subjects, n (%)	Accuracy, n (%)
Abdominal radiography	11 (42)	0 (0)
Abdominal ultrasound	17 (65)	9 (53)
Abdominal CT scan	23 (88)	21 (91)
Colonoscopy	3 (11)	1 (33)

cessfully performed in 4 patients (Fig. 4, 5). Surgical procedures depended on the location of intussusception, the presumed cause, and the viability of intestinal segments.

Preoperative reduction was not adopted in our management of adult intussusceptions. In contrast, intraoperative reduction was generally attempted in all enteric intussusceptions except for 2 patients as malignancy was strongly suspected during surgical exploration. Reduction attempt also failed in 2 patients due to hard irreducible underlying masses. This reduction was intentionally avoided in colonic intussusceptions. However, it was successfully performed in the unique colorectal intussusception in order to reduce the extent of intestinal resection and to preserve the anal sphincter.

The most frequently performed procedure was segmental intestinal resection ($n = 16$, 61%). The other procedures were right hemicolectomy ($n = 6$, 23%), ileocecal resection ($n = 2$, 8%), segmental sigmoidectomy ($n = 1$, 4%), and proctosigmoidectomy ($n = 1$, 4%). All resections were performed with primary anastomosis except for 1 patient who

Fig. 4. Operative view (laparotomy) of an ileo-ileal intussusception (**A**) secondary to adenomyoma acting as a lead point (**B**).

Fig. 5. Operative view (laparoscopy) of an ileo-ileal intussusception.

underwent a stoma for peritonitis caused by intestinal perforation. Details of location, type of intussusception, etiology, and operative procedures are shown in Table 3.

Postoperative complications were observed in 5 patients: 3 cases of surgical site infections, 1 of minor anastomotic leak, and 1 of pulmonary infection. They were successfully treated during the postoperative period.

Location and Etiologies

Surgical exploration confirmed intussusception in all patients and determined its type, location, and causative lesion. Enteric intussusceptions ($n = 19$, 73%) were more predominant than colonic intussusceptions ($n = 7$, 27%). One case of enteric intussusception was jejuno-jejunal, 3 were jejuno-ileal, 12 were ileo-ileal, and 3 were ileocolic. Five cases of colonic intussusception were ileocolic, 1 was colocolonic, and 1 patient had colorectal intussusception. No case of retrograde intussusception was observed in our series.

A pathological lead point was identified in all patients after examination of resected specimens. Benign pathologies were revealed in 17 cases (65%) and malignant processes in 9 cases (35%). The most common benign lesion leading to intussusception was fibrous polyp of Vanek (6 cases). The other benign lesions included lipoma, leiomyoma, adenomyoma, intestinal tuberculosis, Meckel's diverticulum, nonspecific inflammation, and postoperative adhesion. Out of the 9 malignant etiologies, primary adenocarcinoma was the most frequent (4 cases), and others included malignant lymphoma, leiomyosarcoma, gastrointestinal stromal tumor, and undifferentiated carcinoma. Overall, malignant lesions were encountered in 26% of enteric intussusceptions and 57% of colonic ones. Different etiologies are summarized in Table 4.

Discussion

Intussusception in adults is an unusual condition compared to its pediatric counterpart. It is found in only 0.08% of all abdominal surgery [9, 10]. In our study, the mean age was lower than that reported in previous studies [11–13]. The gender prevalence is generally equal between males and females; however, a slight male predominance was recorded in our report [14, 15].

In contrast to the pediatric form, the clinical presentation of adult intussusception is mostly variable and non-specific. Adult patients often present with intermittent or chronic symptoms indicating incomplete intestinal obstruction. The classic triad of abdominal pain, rectal bleeding, and abdominal palpable mass is rarely encountered [16, 17]. Obviously, most of our patients presented with intermittent abdominal pain and nausea/vomiting,

Table 3. Details of location, etiology, and operative procedures of adult intussusceptions

Case	Age, years/ sex	Type	Surgery	Reduction	Location of lead point	Histopathology
1	39/f	jejuno-jejunal	SI segmental resection	Y	jejunum	fibrous polyp
2	48/m	jejuno-ileal	SI segmental resection	N	jejunum	adenocarcinoma
3	26/f	jejuno-ileal	SI segmental resection	Y	jejunum	leiomyosarcoma
4	70/f	jejuno-ileal	SI segmental resection	Y	jejunum	lipoma
5	56/f	ileo-ileal	SI segmental resection (L)	Y	ileum	leiomyoma
6	34/m	ileo-ileal	SI segmental resection	Y	ileum	Meckel's diverticulum
7	56/m	ileo-ileal	SI segmental resection	Y	ileum	B-cell malignant lymphoma
8	25/m	ileo-ileal	SI segmental resection	F	ileum	adenomyoma
9	58/m	ileo-ileal	SI segmental resection	Y	ileum	fibrous polyp
10	21/m	ileo-ileal	SI segmental resection (L)	Y	ileum	postoperative adhesion
11	55/f	ileo-ileal	SI segmental resection	Y	ileum	fibrous polyp
12	70/m	ileo-ileal	SI segmental resection	Y	ileum	fibrous polyp
13	28/m	ileo-ileal	SI segmental resection (L)	Y	ileum	adenomyoma
14	70/m	ileo-ileal	SI segmental resection	Y	ileum	leiomyoma
15	46/m	ileo-ileal	SI segmental resection	N	ileum	GIST
16	51/m	ileo-ileal	SI segmental resection	Y	ileum	undifferentiated carcinoma
17	52/f	ileocolic	right hemicolectomy	F	ileum	nonspecific inflammation
18	22/m	ileocolic	ileocecal resection	Y	ileum	fibrous polyp
19	31/m	ileocolic	ileocecal resection	Y	ileum	fibrous polyp
20	58/f	ileocolic	right hemicolectomy	N	cecum	intestinal tuberculosis
21	47/f	ileocolic	right hemicolectomy	N	cecum	adenocarcinoma
22	30/f	ileocolic	right hemicolectomy (L)	N	cecum	adenocarcinoma
23	32/m	ileocolic	right hemicolectomy	N	colon	Burkitt lymphoma
24	59/f	ileocolic	right hemicolectomy	N	colon	lipoma
25	42/m	colocolonic	sigmoidectomy	N	colon	lipoma
26	32/m	colorectal	proctosigmoidectomy	Y	sigmoid	adenocarcinoma

SI, small intestine; L, laparoscopic surgery; Y, successful reduction; N, reduction not attempted; F, reduction failed; GIST, gastrointestinal stromal tumor.

Table 4. Etiologies and location of adult intussusceptions

Etiologies	Location		Total
	enteric	colonic	
Benign	14	3	17
Fibrous polyp	6	0	6
Lipoma	1	2	3
Leiomyoma	2	0	2
Adenomyoma	2	0	2
Intestinal tuberculosis	0	1	1
Meckel's diverticulum	1	0	1
Nonspecific inflammation	1	0	1
Postoperative adhesion	1	0	1
Malignant	5	4	9
Primary adenocarcinoma	1	3	4
Lymphoma	1	1	2
Leiomyosarcoma	1	0	1
GIST	1	0	1
Undifferentiated carcinoma	1	0	1
Total	19	7	26

GIST, gastrointestinal stromal tumor.

which is consistent with other findings [18, 19]. The presence of weight loss may be associated with serious underlying pathology and should alert surgeons to suspect malignancy [15, 20].

Given its rare prevalence and confusing clinical picture, the diagnosis of adult intussusception is a true challenge and can be missed without imaging studies. Currently, abdominal CT scan is the most effective imaging tool to detect intussusception, with a diagnostic accuracy ranging from 58 to 100% [21, 22]. The typical image is formed by a thickened intestinal wall and mesentery within the lumen giving the pathognomonic “target sign” or “sausage-shaped” appearance according to the axial projection [23–25]. CT scan can identify the exact location of intussusception and reveal any threatening signs of intestinal nonviability. It can also recognize pathological lesions, helping to differentiate intussusception due to organic lesion (lead point intussusception) from intussusception without identified underlying lesion (non-lead point intussusception), which tends to be transient [26, 27]. However, CT scan cannot always distinguish be-

tween a pathological lesion and a thickened edematous intestinal wall except in cases of lipoma [25].

In the hands of experienced operators, ultrasound is a useful technique to detect intestinal invagination. The characteristic images include “target sign” or “doughnut sign” in the cross-section and “pseudo-kidney sign” in the longitudinal section [28, 29]. However, interposition of air contained in distended loops and obesity can affect the image quality and the findings’ accuracy. Consequently, we propose to achieve abdominal CT scan as a regular diagnostic test in all patients admitted for acute abdominal pain or intestinal obstruction.

Colonoscopy is reported to be useful in patients with subacute or chronic intestinal obstruction, especially in colonic forms. It helps to identify parietal lesions and to perform tissue biopsies. Nevertheless, it must be performed in selected patients given the high risk of perforation resulting from parietal ischemia and potentially necrosis [30, 31].

In our study, the preoperative diagnosis rate of intussusceptions was approximately 81%. In the literature, this rate varies between 30 and 90% [32–34]. Our relatively higher rate can be explained by the frequent use of CT scan in our country, particularly in recent years.

The exact mechanism of intussusception is still not well understood. However, most authors report that any lesion in the intestinal wall or intraluminal irritation can alter intestinal peristalsis and induce intestinal invagination [22, 35]. Additionally, adult intussusception is due to an identifiable etiology in nearly 90% of cases [2, 14]. In general, the majority of enteric intussusceptions are secondary to benign lesions. Malignant etiologies represent only 30% of lesions and include primary (mainly adenocarcinoma) or metastatic tumors [16, 36]. Conversely, colonic intussusceptions are more likely due to malignant etiologies in up to 66% of cases [23, 33]. In the present study, enteric intussusceptions were the most frequent type, in line with previous reports [8, 11]. Malignant lesions were more frequently found in colonic intussusceptions than in enteric ones, and the most common malignancy was adenocarcinoma. Other authors reported similar results, confirming the high prevalence of malignant etiologies in colonic intussusceptions [7, 37].

Until now, there is no universally accepted approach to the optimal treatment of adult intussusceptions. Given the large proportion of underlying pathological lesions, surgical management continues to be the mainstay treatment modality for most patients. For colonic intussusceptions, reduction was avoided in our patients. En bloc resection without reduction is recommended by other researchers considering the high rate of malignancy [38–40]. Furthermore, reduction exposes the patient to intraluminal seeding and venous dissemination of tumor cells, intestinal perforation during manipulation with possible

spread of malignant cells into the peritoneal cavity, and increased risk of anastomotic complications in an edematous and inflamed bowel [41, 42].

As malignancy is less common in enteric intussusceptions, reduction can be performed before resection to reduce the extent of intestinal resection and to prevent short bowel syndrome. It is reported that probability of malignancy increases with age, past history of malignancy, and chronic history of Crohn’s or celiac diseases [43–45]. In our study, intraoperative reduction was adopted only for enteric intussusceptions in the absence of suspected signs of malignancy. However, 2 reduced intussusceptions have been shown to be secondary to malignant lesions. This situation must be considered during the surgical procedure because of the inability to distinguish benign from malignant lesions. Similarly, some authors recommended that enteric intussusceptions should also be resected without prior reduction [9, 38]. However, reduction may be attempted only in young patients with strongly suggested benign lesions unless signs of ischemia or strangulation are detected [41, 46].

The expanded use of CT scan has increased the detection of asymptomatic and incidental intussusceptions in adults. These forms, more frequently without a detected pathological lead point, are secondary to transient peristaltic disorders and are likely to be resolved spontaneously. They are designated as idiopathic intussusceptions and usually affect the small bowel rather than the colon [26, 47]. Confronted with this situation, the surgeon is confused about whether the patient must undergo surgical treatment or not. Many authors have recently suggested a more selective approach in the management of enteric intussusceptions. Shenoy [45] proposed that patients presenting with intestinal obstruction, digestive bleeding, abdominal palpable mass, or a pathological lesion on CT scan should undergo surgical treatment. Contrarily, Onkendi et al. [48] reported that enteric intussusceptions without a detectable lead point or obstructive symptoms are probably self-limiting and can be managed nonoperatively. Also, Lvoff et al. [49] and Mateen et al. [50] showed that enteric intussusceptions measuring <3.5 cm in length do not require surgical exploration. However, if signs of intestinal obstruction or parietal ischemia appear during patient monitoring, a surgical exploration is necessary [47, 51]. No idiopathic intussusception was detected in our study, and all cases were secondary to organic lesions. This may be explained by the fact that the data were collected only from the surgical records and CT scan was not systematically performed for abdominal complaints.

Laparoscopic surgery for adult intussusceptions is currently feasible and safe in experienced hands [52, 53]. This approach, with both diagnostic and therapeutic possibilities, offers all the advantages of mini-invasive surgery. It is a useful adjunct to the conservative approach that allows

exploring the abdominal cavity without subjecting the patient to aggressive incisions [51, 54]. Within our institution, laparoscopic activities have increased since 2010 with a young and ambitious surgical team. We have successfully used this approach in 3 cases for segmental intestinal resection and in 1 case of right hemicolectomy with an uneventful postoperative course. Results of surgery in adult intussusceptions are generally good, but the long-term prognosis depends mainly on the underlying diseases [16, 20].

The major limitations of this study were its retrospective design and its small sample size due to the rarity of adult intussusceptions. Inclusion criteria related only to the patients' surgical records were another limitation. Thus, the incidence of this disease is certainly underestimated. A prospective multi-institutional study, involving a larger number of patients, seems to be necessary to draw relevant conclusions.

Conclusion

Adult intussusception is an infrequent pathology with a really challenging diagnosis that requires a high level of suspicion. It should be considered in any patient with subacute abdominal pain. However, the wide use of CT scan, the most effective imaging modality, makes diagnosis easier and more accurate. Adult intussusception is often due to an underlying lesion that requires surgical intervention. Given the high rate of associated malignancy, en bloc resection without attempting reduction should be the treatment of choice of colonic intussusceptions. A more selective approach is recommended for patients

with enteric intussusceptions, especially in asymptomatic and incidental forms. Conservative management with close monitoring can be applied in selected patients.

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Statement of Ethics

The study was performed in accordance with the Medical Ethics Committee's requirements. Given the retrospective design, written consent was not required.

Disclosure Statement

The authors have no conflicts of interest to declare.

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Author Contributions

M.T. participated in the conception and design of the study, performed the collection of data, and drafted the manuscript. A.A.A. contributed to the analysis and interpretation of data and revised the manuscript.

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