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Adult intussusception: a single-center 10-year experience

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Summary
Background Adult intussusception (AI) is a rare condition, usually with a lead point, and for which surgery is the treatment of choice. Given the risks and possible complications of untreated AI, an accurate preoperative diagnosis is of the utmost importance. Although AI remains difficult to diagnose, computerized tomography (CT) is presently considered the best diagnostic tool.

Methods Sixteen patients of 20 years and older with intraoperative diagnosis of intussusception, who underwent surgery between January 2000 and December 2009, were reviewed retrospectively. Patients were assessed concerning clinical presentation, imagiological findings, surgical treatment, and postoperative histological evaluation.

Results Most patients (93.8 %) were admitted via emergency room (ER) due to abdominal pain. Fourteen (87.5 %) AI cases showed an underlying organic cause, e.g., masses or tumors. The most frequent comorbidities were Peutz-Jeghers syndrome (PJS; 18.8 %) and HIV (12.5 %). Eight (50.0 %) intussusceptions were ileocolic and six (37.5 %) were in the small bowel. Total 43.8 % of lesions were malignant. Preoperative diagnosis of intussusception was possible in 50.0 % of cases by ultrasonography (US) and in 81.8 % by CT. US showed no predictive value concerning intussusception location. Total 27.3 % of CTs correctly identified the location, but only 9 % accurately identified the lead point.

Conclusions We propose that all AI cases should be treated with surgical resection without attempting reduction, even when no lead point is detected by imaging studies, and this approach should be based on the oncological criteria. CT can be regarded as the most accurate diagnostic tool for intussusception, although its predictive value concerning location and lead point is still far from ideal.

Keywords: Adult intussusception, Intestinal obstruction, Oncological surgery, CT diagnosis, Emergency surgery

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Introduction

Intussusception occurs when a segment of the gastrointestinal tract (intussusceptum) invaginates (telescopes) into an adjacent segment (intussuscipiens). Adult intussusception (AI) is a rare condition, with most of the cases being pediatric [1]. In children, intussusception is the second most common abdominal emergency after appendicitis [2], being idiopathic in 90 % of cases. In contrast, AI accounts for an estimated 5 % of all intussusceptions and only 1 % of adult bowel obstructions [3]. Despite this low incidence, if not treated, the arterial blood supply to the bowel will be jeopardized, and this can lead to ischemia, perforation, and peritonitis, resulting in a lethal condition [4, 5]. The treatment of choice in children is nonoperative reduction, for example, by rectal air insufflations with surgical reduction only undertaken if this fails. On the contrary, in adults, surgical exploration is the traditional management due to the often malignant nature of a persistent underlying lead point [4, 6–8].

Although preoperative diagnosis of intussusception remains challenging, its detection has become more frequent due to the increased use of ultrasound and computerized tomography (CT). CT is the most effective detection method as it is neither affected by bowel gas, which is common in adults with intussusception, nor bowel obstruction [9]. Most cases detected on CT are not suspected clinically [10, 11].

In the present paper, we retrospectively review 16 cases of AI over a period of 10 years, concerning clinical presentation, therapeutic approach, and anatomopathology, focusing especially on presurgical imagiological findings and their concordance to intraoperative and histological findings.

Materials and methods

Sixteen patients of 20 years and older with intraoperative diagnosis of intussusception, who underwent surgery between January 2000 and December 2009 at Hospital Prof. Doutor Fernando Fonseca E.P.E., Amadora, Portugal, were reviewed retrospectively. Patients were assessed concerning clinical presentation, infection parameters, imagiological findings, surgical treatment, and postoperative histological evaluation. Follow-up information was obtained based on postoperative evaluation during hospital stay and outpatient's visits. All participants provided written informed consent.

Results

Patients

Patient characteristics are summarized in Table 1. Fifteen (93.8 %) patients were admitted via emergency room (ER). Most patients (81.3 %) had a symptom duration of less than 1 month (range 12 h to 1 month). Symptomatology was generally nonspecific: vague abdominal pain was the most common symptom (15 patients, 93.8 %) followed by bowel occlusion (31.3 %) and diarrhea (25.0 %). A palpable abdominal mass was only present in 18.8 % of cases. Seven (43.8 %) patients had known comorbidities: three (18.8 %) Peutz-Jeghers syndrome (PJS), two (12.5 %) HIV, one (6.3 %) a previously operated colon neoplasia, and one (6.3 %) anorexia.

Table 1. Patients characteristics upon admission

Characteristic		Number (%)
Demographics	Age (mean ± SD)	45.2±20.3
	Female	9 (56.3)
	Admitted via ER	15 (93.8)
Signs and symptoms[a]	Abdominal pain	15 (93.8)
	Diarrhea	4 (25.0)
	Palpable abdominal mass	3 (18.8)
	Fever	1 (6.3)
	Chronic constipation	2 (12.5)
	Bowel occlusion	5 (31.3)
	Acute abdomen	2 (12.5)
	Hematochezia	1 (6.3)
Time from symptom onset to surgery	<72 h	6 (37.5)
	3 days to 1 month	7 (43.7)
	1 month to 1 year	2 (12.5)
	>1 year	1 (6.3)
Comorbidities	HIV	2 (12.5)
	Peutz-Jeghers syndrome	3 (18.8)
	Anorexia	1 (6.3)
	Previous surgery to colon neoplasia	1 (6.3)

[a]50 % of patients with compound symptoms
ER emergency room, *SD* standard diviation

Preoperative findings

Preoperative laboratory evaluation showed that the majority of patients (85.7 %) had at least one infection parameter (Table 2). Ten patients underwent abdominal ultrasonography (US) and 11 abdominal CT. US diagnosed five intussusceptions (50.0 %), but only three (30.0 %) suggested their location, and none referred a lead point. CT diagnosed eight (72.7 %) intussusceptions, with 63.6 % of CTs suggesting their location and 18.2 % a lead point.

Intraoperative findings of intussusception surgery

Total 50.0 % of intussusceptions were ileocolic and 37.5 % were in the small bowel. One patient (6.3 %) had a colocolic intussusception and one a colorectal—Table 3.

All patients underwent surgical treatment. The most common surgical procedure was segmental small bowel resection (35.3 %), followed by ileocolic resection and right hemicolectomy (29.4 and 23.5 %, respectively).

Table 2. Preoperative findings

Parameter	Number (%)	Mean (SD)	Range
Laboratory evaluation			
Leukocytosis (cells/mm^3)	9 (56.3)	13.911 (3.298)	10.000–21.200
Neutrophilia (%)	7 (43.8)	86.7 (6.1)	80.0–93.0
PCR>1.0 (mg/dL)	8 (50.0)	4.3 (3.8)	1.1–13.4
Imagiological findings			
Abdominal US	10 (62.5)	–	–
Intussusception diagnosis	5 (50.0)	–	–
Intussusception location	3 (30.0)	–	–
Reference to lead point	0 (0.0)	–	–
Abdominal CT	11 (68.8)	–	–
Intussusception diagnosis	8 (72.7)	–	–
Intussusception location	7 (63.6)	–	–
Reference to lead point	2 (18.2)	–	–

CT computerized tomography, *SD* standard diviation, *US* ultrasonography

Table 3. Intraoperative findings of intussusception surgery

Parameter	Number (%)
Surgical procedure	
Patients admitted via ER	
Segmentar small bowel resection	6 (35.3)[a]
Ileocolic resection	5 (29.4)[a]
Right hemicolectomy	4 (23.5)
Hartmann's procedure	1 (5.9)
Elective	
Left hemicolectomy with rectopexy	1 (5.9)
Type of intussusception	
Ileocolic	8 (50.0)
Enteroenteric	6 (37.5)
Colocolic	1 (6.3)
Colorectal (elective patient)	1 (6.3)

[a]One patient with both
ER emergency room

Of the four ileocolic resections, one was a relapse after 3 years of a right colectomy due to colon carcinoma. The other three were in patients with intraoperative confirmation of terminal ileon tumors, with histological diagnosis of adenocarcinoma, Kaposi sarcoma, and mucosa-associated lymphoid tissue (MALT) lymphoma.

Postoperative course

Of the 16 patients, 4 (25 %) had some kind of postoperative complication, 2 (12.5 %) infection of the surgical incision, 1 (6.3 %) hemoperitoneum and shock after 24 h of segmental jejunal resection resolved by emergency surgery with ligation of a bleeding vessel in the mesentery, and 1 (6.3 %) occlusion by bridles after 1 year resolved by resection. The remaining 12 patients showed no postoperative complications. Mean time from surgery to discharge was 6.8±1.5 days.

Diagnosis

Table 4 summarizes preoperative imaging results, intraoperative findings, and final histological diagnosis. US showed a diagnostic accuracy of 50.0 %, but no predictive value concerning intussusception location or lead point. The overall predictive diagnostic value of CT was 81.8 % with 27.3 % of CTs correctly identifying the intussusception location, but only 9 % accurately identifying the lead point. Seven (43.8 %) intussusceptions were histologically confirmed to be of malignant etiology—one GIST, two MALT lymphomas, and one Kaposi sarcoma. The other three were adenocarcinomas, one in the terminal ileon and the other two colon carcinomas. Of these seven malignant neoplasms, two MALT lymphoma and one Kaposi sarcoma, were identified in the two HIV-positive patients.

Table 4. Pre- and intraoperative intussusception location and histological diagnosis

Location by US	Location by CT	Intraoperative	Histological diagnosis
Sigmoid	NP	Ileocolic	Adenocarcinoma
Left colon	NP	Ileoileal + jejunojejunal	Hamartomatous polyp (PJS)
Right colon	NP	Ileocolic	Hyperplastic polyp
NP	No	Enteroenteric	Hamartomatous polyp (PJS)
NP	Ileocolic	Ileocolic	Lipoma
NP	No	Ileoileal	Meckel diverticulum
NP	Colocolic	Ileocolic	Adenocarcinoma
NP	Recto-sigmoid junction	Rectosigmoid junction	Vilous adenoma with low-degree displasia
NP	NP	Colorectal	Unspecific inflammation; melanosis
No	No	Enteroenteric	Hamartomatous polyp (PJS)
No	Enteroenteric	Jejunoileal	MALT
No	Right colocolic	Ileocolic	MALT
No	Ileoileal	Ileocolic	Kaposi
No	Colocolic	Jejunojejunal	GIST
No	Ileocolic	Ileocolic	Unspecific inflammation
No	No	Ileocolic	Adenocarcinoma

Overall percent concordance by US = 0.0 % and by CT = 27.3 %
CT computerized tomography, *GIST* gastrointestinal stromal tumor, *MALT* mucosa-associated lymphoid tissue, *NP* not performed, *PJS* Peutz-Jeghers syndrome, *US* ultrasonography

Follow-up

All patients continued to attend the outpatient clinic. The number of days until discharge varied between 5 and 11 with a mean of 6.8±0.4 days from surgery to discharge. Postoperative follow-up varied between 1 month and 120 months, with a mean of 39.7±10.6 months. Three (42.9 %) of the patients with malignant tumors, 1 GIST, 1 MALT, and 1 Kaposi, died within less than 5 months after surgery, due to underlying disease progression—metastatic melanoma in one patient and AIDS in two other patients. One patient with a histological diagnosis of adenoma died 4.5 years after surgery from an unrelated cause.

Discussion

AIs are traditionally thought to have an identifiable cause in the majority of cases [4]. Although the mechanism of intussusception development is yet unknown, a variety of factors can act as a lead point and, theoretically, any process that alters normal peristalsis or any lesion in the intestinal wall or irritant within the lumen, which alters

normal peristalsis, is able to initiate an invagination [12], led on by edema, inflammation, or infection [13]. Meckel's diverticulum [4], inflammation in HIV-positive patients [14], malabsorption syndromes [15], and gastrointestinal infections have all been associated with intussusception. Intussusception has also been related with severe hyperglycemia [16] and acute appendicitis in young adults [17] and has been reported in pregnancy [18]. Increasing evidence also suggests an association with a roux-en-Y bypass in gastric bypass patients [19, 20]. In our study, at the time of admission, seven patients (43.8 %) had diagnosed associated pathologies which could be responsible for the invagination, three (18.8 %) with PJS, and two (12.5 %) HIV positive. Although several reports identify these two pathologies as the underlying cause of AI [14, 21–23], others mention this only occurs rarely [24, 25]. Given an overall 31.3 % of the studied patients had either PJS or HIV; our results support these two pathologies as a frequent cause of AI.

Intussusceptions with a lead point may result in abdominal pain, nausea, vomiting and, occasionally, obstructive symptoms [13, 26]. If the intussusception is a result of an underlying malignancy, patients may also report weight loss [13]. Symptoms are in most cases of long duration, varying from several weeks to months [9], although the occasional acute pain may occur. Patients without an identifiable lead point may experience symptoms such as abdominal pain with cramping [27]. The findings of this study are in accordance with these reports, given all patients admitted to the ER complained of severe abdominal pain, with a history of vomiting in 50.0 % (n=8) of cases, and associated intestinal obstruction (31.3 %) and diarrhea (25.0 %). Six patients (37.5 %) had an acute course of symptoms of less than 72 h, with the remaining patients showing subacute courses of weeks or months, with symptom remissions and intensifications. According to Warshauer and Lee [28], only in 21.2 % of intussusceptions the physical examination detects the presence of an abdominal mass, which is in accordance with the observed three (18.8 %) patients in this study. Also, the majority of patients (85.7 %) had at least one infection parameter, which supports infection as a common comorbidity in intussusception.

Most adult small bowel intussusceptions are secondary to benign lesions [29], but in the colon they are more likely to be associated with a malignant etiology [4, 6–8]. Given this high incidence of malignant tumors as the lead point in colon intussusception, treatment usually consists of surgical resection and reduction is not attempted due to the risk of perforation and potential spillage of microorganisms and malignant cells [4]. However, in small bowel intussusception with obstruction or severe symptoms, a reduction is attempted unless ischemia or strangulation is detected [4].

Recently, with the development and more frequent use of CT, asymptomatic cases have been presented where surgical approach is not necessary. Given this increased detection of intussusception, even in asymptomatic patients, and given the recognition that some may be transient, there is an ongoing controversy regarding the optimal management of these cases. The general consensus is that proximal small-bowel intussusceptions measuring less than 3.5 cm in length and without obstruction or an obvious lead point are likely to be self-limiting and of no clinical significance [30], and larger sections should be monitored conservatively [31], only resorting to surgery if complications arise.

In our study, treatment of choice for all patients was surgery, with nearly all cases (93.8 %, n=15) requiring emergency surgery, as previously reported [32], and 37.5 % (n=6) undergoing segmental small bowel resection. Although there is no consensus regarding the optimal surgical approach, and there is still controversy about reduction before resection [32], several authors suggest resection as the best treatment option in adults, given nearly half of colonic and enteric intussusceptions are associated with malignancies [3, 33]. Moreover, due to the several risks and severe complications associated with intussusceptions and their delayed treatment such as intraluminal seeding and dissemination, perforation and peritoneal dissemination with edematous, and fragile bowel wall [34, 35], most surgeons recommend surgery regardless of the nature of their cause [36, 37].

Our results show that 46.7 % of all patients with intestinal intussusception are admitted to the ER with acute abdomen or bowel occlusion and should undergo emergency surgery, which is not a consensual recommendation [5, 38]. Fourteen (87.5 %) AI cases had an underlying organic cause, e.g., masses or tumors, which cannot be histologically diagnosed in the ER. Only 2 cases (12.5 %) showed chronic unspecific inflammation without endoluminal mass. Therefore, the chance of a mechanical obstruction is very high, and frequently undetected by US or CT. Moreover, 7 of the 16 cases were histologically confirmed to be malignant neoplasms, in our opinion with a clear surgical resection indication, since they presented with obstruction or acute abdomen. Given these results, and since reduction of the intussusception will not resolve the underlying pathology, we propose that all AI cases should be treated with surgical resection without attempting reduction, even when no lead point is detected by imaging studies, and that this surgical approach should be based on oncological criteria.

Our study did not include any case of laparoscopic surgery given the lack of experience in colorectal laparoscopic surgery in the ER 10 years ago. However, presently, the potential of laparoscopic surgery is a reality, namely for differential diagnosis and potential identification of lead point and, should it be considered necessary, then proceed to surgical resection based on oncological criteria. This laparoscopic approach has all the diagnostic and therapeutic advantages of mini-invasive surgery, namely a less traumatic and swifter postoperative recovery, with a lower rate of surgical complications. However, a laparoscopic approach to acute abdomen, and particularly intestinal occlusion, is far from consensus, having been amply debated at the 13th European Congress of Trauma and Emergency Surgery, held in Basel on May 2012.

Intussusception can occur anywhere along the gastrointestinal tract, and may be classified according to location—enteroenteric, ileocolic, or colocolic—or etiology—neoplastic (benign or malignant), nonneoplastic, or idiopathic [9, 10]. Gastric intussusception is very rare, whereas enteroenteric intussusception (within the small and large bowels) is more common, accounting for more than 40 % of cases [15]. In the present study, the most frequent locations were ileocolic (50.0 %, $n=8$) and enteroenteric (37.5 %, $n=6$), among which 43.8 % ($n=7$) histologically confirmed to be of malignant etiology. Of the seven malignant neoplasms, two MALT lymphoma and one Kaposi sarcoma, were identified in the two HIV-positive patients. In fact, these patients are considered at risk for intussusception, as a result of their high susceptibility for recurrent infection and inflammation, as well as tumors such as Kaposi sarcoma [14]. According to Wood et al. [39], an intussusception diagnosis should be considered likely to occur in HIV-positive patients presenting with abdominal pain.

Although there is an ongoing controversy regarding the best diagnostic means for intussusception, imaging techniques are considered by many as the most reliable diagnostic tools. Abdominal US and CT are the most commonly used techniques, but the advantages and disadvantages of each method are still debated. US is useful for obstruction assessment purposes, and may, in some cases, identify its cause [40]. However, diagnosis may be difficult, given it can be masked by the presence of intestinal gases [4]. CT can assess both the nature and location of tumors, if present, as well as identify soft tissues involved in the invagination, obstruction, and threatening signs of bowel viability [36], with the added advantage of not being affected by bowel gas, common in adults with intussusception [9], being currently considered the most sensitive radiologic method to confirm intussusception, with a reported diagnostic accuracy of 58–100 % [35, 41]. In the present study, 62.5 % ($n=10$) of cases underwent US, resulting in five diagnosis of intussusception, but only three suggesting its location as sigmoid, right and left colon, without reference to lead point. Intraoperatively, these three cases were confirmed as ileocolic, ileoileal, and ileocolic, respectively. Therefore, and although the diagnostic accuracy was of 50.0 %, US showed no predictive value concerning intussusception location. Additional CT was performed in seven of these patients, of which six (85.7 %) were predictive of intussusception diagnosis and five (71.4 %) of its location, without reference to lead point. Intraoperatively and histologically, two (40 %) of the locations were confirmed, one enteroenteric and one ileocolic. The other three, identified as right colocolic, enteroenteric, and colocolic by the CT, were histologically diagnosed as ileocolic, ileocolic, and enteroenteric, respectively. Of the four patients who underwent CT without prior US, two were diagnosed including location and lead point. Of these two, one was an ileocolic intussusception due to a lipoma in the right colon, which was confirmed intraoperatively. The other, identified by CT as a colocolic intussusception due to neoplasia of the hepatic angle, was intraoperatively and histologically diagnosed as an ileocolic intussusception due to adenocarcinoma in the adenoma of the terminal ileum. Therefore, the overall predictive diagnostic value of CT was 81.8 %, which is in accordance with previous reports [12]. Total 27.3 % of CTs correctly identified the intussusception location, but only 9 % accurately identified the lead point.

These results suggest that US or CT are not as sensitive and specific as widely considered for the diagnosis of abdominal pathologies requiring emergency surgery, and support the recommendation of surgical resection in AI, even when no lead point is detected.

Conclusions

AI is a rare condition, often being the first symptom of an underlying tumor. Surgical approach should be based on oncological criteria, and resection without attempting reduction should be the preferred choice. In patients with a history of polypoid syndromes such as PJS, or HIV with bowel obstruction, an intussusception diagnosis should always be suspected and further investigated. CT can be regarded as the most accurate diagnostic tool for intussusception, although its predictive value concerning location and lead point may still be considered far from ideal. Being an unusual and challenging condition, requiring an accurate preoperative diagnosis in order to expedite immediate surgical therapy, as a missed initial diagnosis may delay appropriate treatment and cause serious complications, it is of the utmost importance to have a thorough knowledge of the underlying lead point, and only improving imaging techniques will enable us to reach a concise diagnosis. Although more research in the field is warranted in order to ascertain the best diagnostic and most successful therapeutic approaches, the authors propose that the surgical approach to AI should be based on oncological criteria: an AI should be assumed to have an underlying oncological cause and should surgically resected without attempting reduction, even when no lead point is detected by imaging studies.

Conflict of interest
With regards to the original paper entitled "10 years of adult intussusception cases", submitted to *European Surgery*, the authors declare that there is no conflict of interest. No funding was received to support this work.

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