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Case report

Adenocarcinoma of the sigmoid colon causing sigmoido-rectal intussusception: A rare entity in adults

Michele Fiordaliso [a,*], Flavia Antonia De Marco [b], Raffaele Costantini [c], Paolo Panaccio [d], Pierluigi Lelli Chiesa [e]

[a] Department of Surgery, Erbach General Hospital, Germany
[b] Klinikum für Innere Medizin Friedrichshafen, Germany
[c] Institute of Surgical Pathology, Department of Medical, Oral and Biotechnological Sciences, University of Chieti, Italy
[d] General Surgery Unit, Renzetti Hospital, Lanciano, Italy
[e] Pediatric Surgery Unit, Hospital "Santo Spirito" of Pescara and University "G. d'Annunzio" of Chieti Pescara, Pescara, Italy

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ABSTRACT

Introduction: Intussusception occurs when a more proximal portion of the bowel (intussusceptum) invaginates into the more distal bowel (intussuscipiens). The pathomechanism is thought to involve altered bowel peristalsis at the intraluminal lesion, which is then a lead point for the intussusceptum. Intestinal intussusception is rare in adults, accounting for approximately 1 % of all bowel obstructions. We report a unique case in which a partially obstructing sigmoid cancer caused full thickness rectal prolapse requiring surgical intervention.

Presentation of case: A 75-year-old male presented in the emergency department due to anal haemorrhage for 5 days. On clinical examination his abdomen was distended with signs of peritoneal irritation in the right quadrants. The CT scan showed sigmoid-rectal intussusception with an sigmoid colonic tumour. The patient underwent emergency anterior resection of the rectum without reduction of the intussusception. Histological examination revealed a sigmoid adenocarcinoma.

Discussion: Intussusception is the most common urgent situation among the pediatric population but its incidence in adults is very rare. The diagnosis is difficult to establish with history and physical exam findings alone. Since in adults, unlike children, in most cases a malignant pathology acts as a lead point, the treatment of this pathology still reserves doubts. Recognizing and understanding pertinent signs, symptoms, and imaging findings is essential to the early diagnosis and appropriate management of adult intussusception.

Conclusion: The appropriate management of adult intussusception is not always clear cut. There is controversy about the reduction before resection in cases of sigmoidorectal intussusception.

1. Introduction

Although intussusception is a common condition in children, it is rare in adults. Adult intussusception differs from pediatric intussusception in various respects, including etiology and clinical characteristics. Intussusceptions are classified according to the intestinal tract involved in four categories: enteric, ileocolic, ileocecal, and colic. In contrast to childhood intussusception, in adults it is associated with malignant lesions, particularly in the large bowel rather than in the small bowel. Its preoperative diagnosis and treatment in adults is difficult because of nonspecific abdominal symptom and because it rarely presents with the classic triad of vomiting, abdominal pain and passage of blood per rectum. An intussusception that involved only the jejunum or ileum was considered an enteric intussusception. An intussusception that involved the ileum and the colon was designated an ileocolic intussusception [1]. An intussusception that involved only the colon was considered a colocolonic intussusception and one that involved the sigmoid colon and rectum was considered a sigmoidorectal intussusception. A proximal segment of the bowel telescoped into the lumen of the adjacent distal segment was defined as antegrade intussusception. A distal segment of the bowel telescoped into the lumen of the adjacent proximal segment was defined as retrograde intussusception. In our review we only considered colocolonic and discusses the optimal preoperative diagnosis and surgical management techniques. Sigmoido-rectal intussusception is

* Corresponding author at: Department of General Surgery, Albert-Schweitzer-Straße 10-20, 64711 Erbach, Germany.
E-mail addresses: michele.fiordaliso@gmail.com (M. Fiordaliso), r.costantini@unich.it (R. Costantini).

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a subtype of intestinal intussusception in which an intussusceptum composed of distal sigmoid or rectum prolapses through the anal canal. This case report has been reported in line with the SCARE 2020 criteria.

2. Materials and methods

Here, we present and discuss a case of Sigmoid-rectal intussusception in adults. In addition, a search of the English-language medical literature using PubMed and Google Scholar was conducted for articles related to gastrointestinal intussusception; the key words used were intussusception in adults, tumour Sigmoid-rectal intussusception and colocolonic intussusception. If there were any missing data, the corresponding authors of the articles in question were contacted by email. Articles containing adequate information, such as publication year, patient age, sex, duration of complaint, radiological tools, presence of palpable mass, surgical approach, were included, while studies and comment articles with insufficient clinical and demographic data were excluded.

3. Case presentation

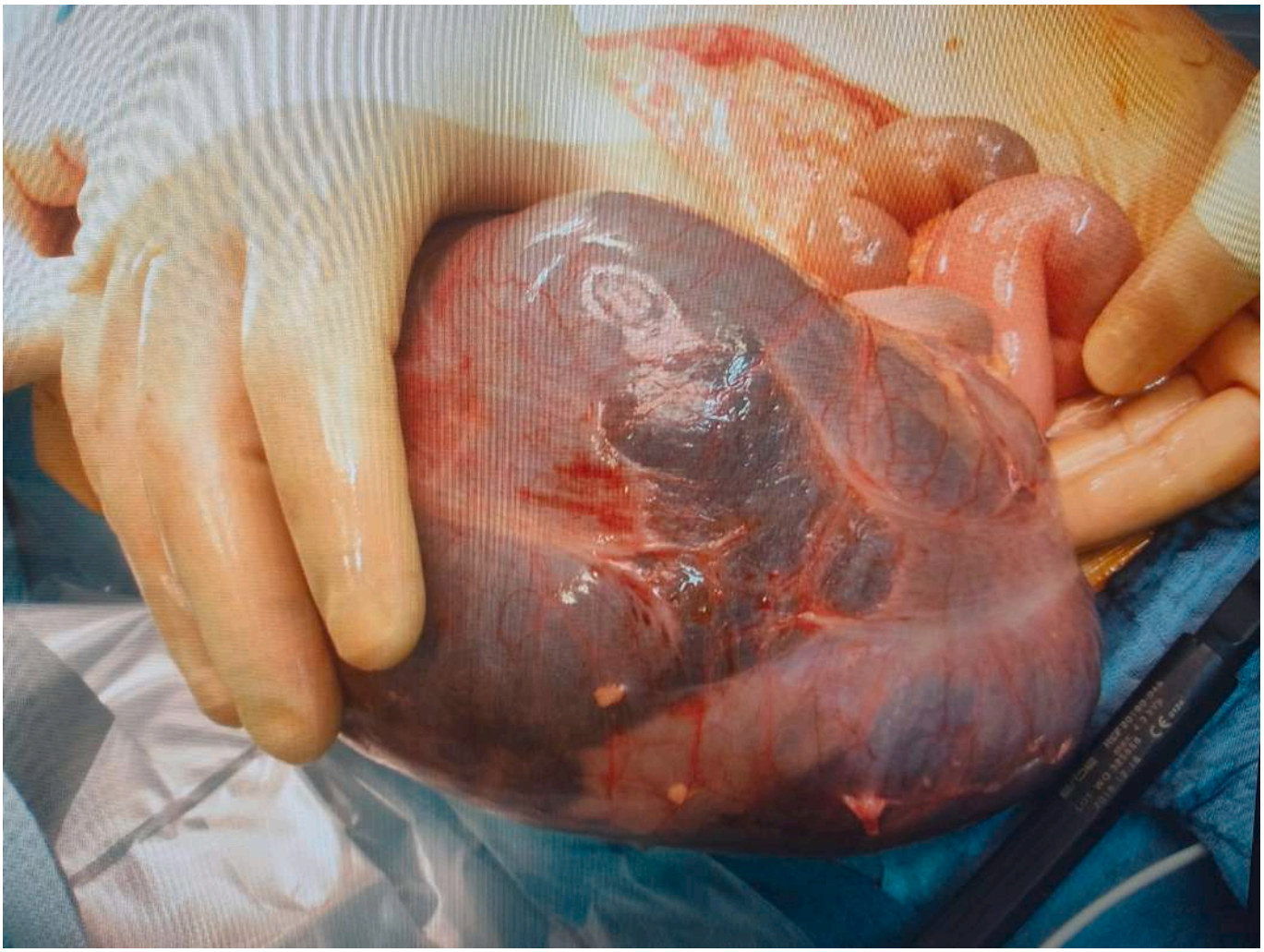
We present the case of a 75-year-old man admitted to our hospital due to anal haemorrhage for 5 days. At the time of admission, his temperature was 38.5 °C and he showed signs of shock: he was tachycardic to 110 beats per minute, and with low blood pressure of 80/60 mmHg. On clinical examination his abdomen was distended with signs of peritoneal irritation in the right quadrants. The bowel was closed to faeces and gas. On rectal examination a hard swelling was evident about 7 cm from the anal rima. Unfortunately, it was not possible to appreciate the implant base, as if it had been a 'pedunculated polyp'. The anal sphincter was normotonic. The white blood cell count was 20.000 cells/mm^3 and C-reactive protein level was 250 mg/L. An X-ray of the abdomen (Fig. 1) showed a distended colon. The CT scan showed sigmoid-rectal intussusception (Fig. 2) with an approximately 4 cm sized sigmoid colonic tumour with massive distension of the cecum with a maximum diameter of 10 cm. The patient underwent emergency anterior resection of the rectum without reduction of the intussusception and right haemicolectomy with ileotransverse Anastomosis due to ischaemia of the cecum caused by prolonged distension (Figs. 3–4). The two ends of the colon were very edematous and also considering the non-congruence of the caliber of the two lumens, no anastomosis was performed between the descending colon and the rectum. A descending colostomy was constructed in the left lower abdomen, and the abdominal incision was closed with several drainage tubes into the abdomen. The patient was treated with 7 days of metronidazole, and ceftriaxone. Improving leukocytosis and decreasing CRP occurred in the following days. His symptoms improved and he tolerated the diet well. We started a liquid diet on the first postoperative day and phases in soft blend diet and normal regular diet after confirming the passage of flatus through the colostomy. The postoperative course was normal and the patient was discharged 10 days after the operation. Histopathological examination showed a localised adenocarcinoma in the distal sigmoid colon with lymph node metastases (T3N2aM0).

Fig. 1. Abdominal x-ray showing intestinal obstruction with massive distension of the right colon.

Fig. 2. Axial computed tomography image of sigmoido-rectal intussusception with voluminous tumour seen within the sigmoido lumen.

4. Discussion

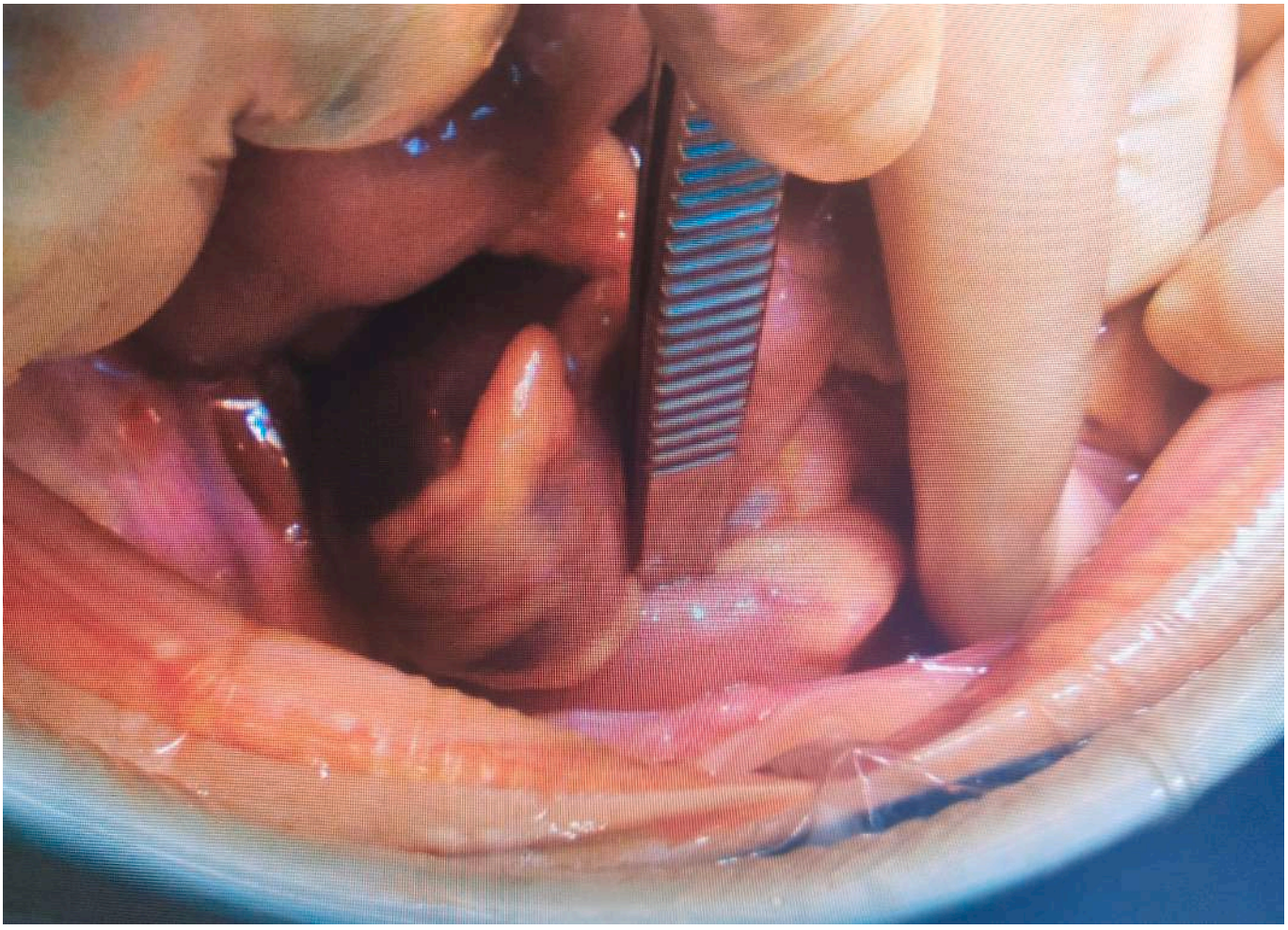
Intussusception is the most common urgent situation among the pediatric population under the age of 3 years, but its incidence in adults is very rare, accounting for only 5 % of all intussusception occasions. Age of presentation in adulthood varies with a mean age of 45 years [2] and median age of 70 years. In adults, small bowel intussusception is more common than colonic, and is triggered by a neoplastic lead point in 66 %–75 % [3]. Almost 50 % of the lesions are benign such as lipomas, lymphoid hyperplasia, leiomyomas, hemangiomas and polyps. Significant percentage of small bowel intussusception may be malignant in the origin, including carcinoma, lymphoma and gastrointestinal tumour [4]. Moreover, most tumours that cause small bowel intussusception represents metastatic disease [5]. Other rare causes include infections, adhesions, inflammatory bowel disease (IBD) granulomas and Meckel diverticulum. Colocolonic intussusceptions are extremely rare, whereas >80 % are associated to malignant aetiologies [6]. Intussusception in adults presents with nonspecific gastrointestinal symptoms such as nausea, vomiting, abdominal pain, bloody bowel movements, or constipation. Cases in whom organic lesions act as the lead point classically present as bowel obstruction. Unlike children, in whom abdominal pain or a palpable abdominal mass predominates, symptoms in adults are usually non-specific, periodical, or even asymptomatic [2,5,7]. For pediatric patients, ultrasonography is commonly used as a cost-effective and non-invasive means of identifying a target sign indicating intestinal intussusception. However, its utility is compromised by air within



Figs. 3, 4. Intraoperative images showing massive distension of the right colon with necrosis of the wall and Sigmoid rectal intussusception (See the surgical instrument inserted between the two invaginated cylinders).

the bowel lumen, which hinders transmission of ultrasonic waves. In addition, it may be difficult to interpret abdominal ultrasound findings in adults with thick abdominal walls and greater distance between the skin and target anatomy. Hydrostatic reduction under radiologic control, as employed in children, is less effective in adults and risk of perforation during the procedure is not negligible [8]. The most sensitive diagnostic test for this disease is CT, which can identify location, surrounding tissue, and presence or absence of a lead point. Since adults who present with intussusception often do have a lead point that can cause obstruction and ischemia, surgery is classically the primary treatment. However, cases without a physical lead point usually resolve spontaneously [9]. In pathogenesis of intussusception, peristalsis and ingested food push the lesion with the adjacent bowel, which telescopes into the relaxed intestinal segment distal to it [10,11]. Colonoscopy provides direct visualization of the intussuseptum and associated intraluminal pathology and allows for biopsy and tissue diagnosis. Differentiating between benign and malignant underlying pathology informs decision-making regarding operative resection, an area of controversy.

We report below some cases of colonic intussusception and their respective lead-point treatments. Maldonado et al. [12] in their review report the case of a 27-year-old patient, recently diagnosed with ulcerative colitis and giant pseudopolypus, who had intussusception of the colon at the splenic flexure with subsequent intestinal obstruction. CT imaging confirmed the diagnosis and the patient underwent resection of the invaginated colon tract, without any bowel reduction. Draganic et al. [13], on the other hand, reported a case of colon invagination in a patient with Crohn's disease, which was treated conservatively with contrast enema and without any recurrence at a 6-month follow-up. Subsequent endoscopy showed that the point of derivation was probably a previous polypectomy site in the sigma [14]. In patients with IBD, invagination is a rare event. In cases where it results from giant pseudopolyp or oedematous mucosa following polypectomy, some authors have proposed conservative management, making surgical resection necessary only in cases of intestinal obstruction or failure of conservative methods [15,16]. Kasuga et al. [17] describe the case of a patient with colon invagination, caused by a metastatic melanoma, for which endoscopic reduction was performed and surgery was postponed as an elective. This was a 64-year-old patient who had undergone multiple surgeries due to a melanoma and was currently undergoing chemo and immunotherapy. During the latter, he reported abdominal pain associated with diarrhoea and bloody stools. Physical examination and laboratory tests did not reveal any findings warranting emergency surgery. Computed tomography showed intussusception at the level of the descending colon without signs of necrosis and intestinal perforation. The intussusception was reduced endoscopically. De Figueiredo et al. [18] show a rare case of transverse colonic intussusception caused by a large lipoma. The abdominal CT scan showed an invagination of the



Figs. 3, 4. (*continued*).

colon with partial obstruction of the lumen, due to an intraluminal lipomatous mass of about 6 cm. As the patient showed no signs of intestinal obstruction, conservative therapy was performed and endoscopic examination was performed after three days. The colonoscopy showed a mass of fibroelastic consistency compatible with a lipomatous mass. Due to the presence of an area of necrotic tissue in the mucosa, any attempt at reduction was avoided because of the risk of perforation. Subsequently, the patient underwent laparoscopic colic resection of the affected tract, without any reduction manoeuvre. The management of intraluminal lipomatous lesions of the colon is traditionally surgical and allows selective resection, depending on the length of the invagination and the extent of inflammation. In the case described above, his chronic condition and the absence of signs of intestinal obstruction allowed surgery to be postponed [19]. Tatsuta et al. [20] reported the case of a 34-year-old patient who presented with sudden abdominal pain. Computed tomography revealed an intussusception of the transverse colon in the absence of intestinal obstruction, bowel wall oedema or tumour. Due to severe abdominal pain, diagnostic laparoscopy was performed. There were no signs of perforation, ischaemia of the intestinal wall or tumour in the abdominal cavity. A diagnosis of idiopathic anterograde colo-colonic invagination was made. Laparoscopic surgical reduction was achieved by combining gentle direct pressure on the anal side of the transverse colon and gentle traction on the oral side. Due to oedema of the intestinal wall a partial resection of the intussuscepted bowel was performed. No tumour was evident on macroscopic examination. Histopathological examination showed oedema and vasodilatation of the submucosa.

Normally in adults Surgical reduction of the intestinal invagination prior to resection is not recommended because of the risk of intestinal perforation. Furthermore, if the intussusception is associated with a malignant tumour, reduction may cause dissemination of tumour cells. In contrast, the utility of laparoscopic surgical reduction in children with intestinal invagination is widely recognised, as most pediatric cases are idiopathic [21].

Since not all cases of adult intestinal invagination are associated with cancer, laparoscopic surgical reduction is feasible if idiopathic intestinal invagination is confirmed by preoperative imaging, as illustrated in the case described. The reduction success rate by laparoscopy was $\geq$70 %, and the success rate was particularly high in ileo-colonic intussusception [22]. The reduction enables avoiding the excessive length of the bowel resection. Surgical reduction methods for adult intussusception are not definitive. These methods involving a combination of delicate direct pressure on the anal side of transverse colon and gentle pulling on the oral side, which is often used for children [23], may be applicable to adults as in the present case. For patients with sigmoidorectal intussusception due to a malignant tumour, the decision on the operative procedure may be modified according to the involvement of the lower rectum. If the latter is infiltrated, an abdominoperineal resection is indicated. However, without evidence of distal disease, an initial reduction may allow sphincter salvage as advocated by Matsuda et al. [24]. In this sense, we would like to emphasise the significance of attempted manual/laparoscopic reduction of the sigmoidorectal intussusception prior to resection, especially for those cases caused by benign lesion for which an abdominoperineal resection would be unnecessary.

5. Conclusion

The appropriate management of adult intussusception is not always clear cut. Most authors agree that operative management of adult intussusception is almost always indicated because of high likelihood of neoplasm as a leading point, particularly if this occurs in the colon. There is controversy about the reduction before resection in cases of sigmoidorectal intussusception, although most surgeons agree that primary surgical resection without a prior attempt at reduction is the treatment of choice in colocolic adult intussusception [5,25,26]. Several aspects to consider for reduction prior to resection are the reduction of externally viable bowel despite mucosal necrosis, intraluminal or transperitoneal seeding, venous embolization of malignant cells, and spillage of succus through inadvertent perforation [5]. In patients with a long segment of invaginated bowel, reduction may be attempted to avoid extensive bowel resection. In particular, in colon intussusceptions with long invaginated segments, intraoperative colonoscopy may be useful to exclude a malignant lesion and thus proceed with an attempt at reduction [27]. In patients in whom a benign polypoid mass is suspected, an initial reduction followed by enterotomy and polypectomy may be attempted [28]. Reduction can be performed manually, but cases of laparoscopic reduction have also been described.

6. SCARE checklist

The work has been reported in line with the SCARE checklist [29].

Ethical approval

This is a Case Report for which the patient provided written informed consent.

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CRediT authorship contribution statement

Michele Fiordaliso: Study design, data collection, writing.
Raffaele Costantini: Data collection, writing.
Flavia Antonia de marco: Data collection, writing.
Paolo Panaccio: Data collection, writing.
Pierluigi Lelli Chiesa: Study design.
All authors have approved the final article.

Guarantor

Fiordaliso Michele

Research registration number

Not applicable.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in Chief of this journal on request.

Declaration of competing interest

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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