Adult intestinal intussusception: a case report of a rare clinical entity

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Riassunto

L'INVAGINAZIONE INTESTINALE NELL'ADULTO. CASO CLINICO DI RARO RISCONTRO

L'invaginazione intestinale nell'adulto è un'entità clinica rara. Essa obbliga il chirurgo a prendere in considerazione una vasta gamma di fattori etiologici e di scegliere opportunamente la strategia terapeutica. L'interrogativo principale in questi casi è, infatti, se sottoporre il paziente a resezione intestinale oppure no.
Gli AA, prendendo spunto da un caso di invaginazione intestinale in una giovane donna con storia di sindrome di Peutz-Jeghers giunto alla loro osservazione, rivedono la letteratura e riportano la loro esperienza.
Parole chiave: Intussuscezione, Peutz-Jeghers, ostruzione intestinale.

Abstract

Intussusception in adult is a rare entity that challenges the surgeon opening a wide range of issues in order to define the etiology and therapeutic strategy.
Whether to resect or not the bowel is the main question. The answer can be given only after having seen the site of obstruction and the etiology. Colonic intussusception is best treated by resection. Also small bowel intussusception can require resection if a neoplasm is the cause. Peutz Jeghers can be one of these causes as is seen in the case we report.
Key words: Intussusception, Peutz-Jeghers, bowel obstruction.

Introduction

Intestinal intussusception is a rare cause of intestinal obstruction in adults and few general surgeons will see more than one or two of such cases during their career (5-11). The intussusception is more frequently observed in the pediatric population. The clinical entity observed in children differs greatly in etiology from the one seen in the adults. Furthermore the classical clinical presentation is rarely seen (1).
Although the nature and the location of the intestinal obstruction secondary to intestinal intussusception may not always be easily predicted, very little is often required in order to make diagnosis of bowel obstruction and treat the patient properly (1-4).
A young female patient was recently referred to us with bowel obstruction due to intestinal intussusception. She offered us the opportunity to review the topic and the issues related to the management of this condition.

Case report

F.M., is a 49 year old, thin, healthy, female with past medical history remarkable only for Peutz-Jeghers syndrome diagnosed about 10 years earlier.
She was now complaining of a new, right sided, coliky, abdominal pain, experienced throughout a full week. This pain was without irradiation and accompanied by nausea, occasional vomiting, obstipation, and a visibile mass of the right flank.
Symptoms had sudden onset, several hours duration and usually subsided with pain medications. She never had any of those symptoms before in her life.
A thorough work-up done years earlier, showed multiple, amartomatous, polyps scattered from the stomach, throughout the duodenum to the colon. The small bowel appeared to be crowded with dozens of polyps.

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On physical exam the patient was afebrile, hemodynamically stable but dehydrated complaining of crampy abdominal pain. She was thin and the mildly dolent, right lower quadrant mass was easily visible and palpable.
There was not guarding or rebound and bowel sounds were present.
Rectal exam was unremarkable, heme negative.
Plain films of the abdomen on admission did not show air-fluid levels, but rather a paucity of gas with a questionable mass.
Ultrasound showed a gas empty, sausage-shaped mass.
Electrolyte imbalances, mild elevation of plasma creatinine and signs of chronic anemia were the most relevant lab value changes.
After the appropriate electrolytes replacement and rehydration the patient was taken to the operating room.
Intraoperative findings were consistent with an intussuscepted terminal ileum, telescoped half way into the right colon.
Exploration of the reminder small bowel revealed numerous intraluminal digiunal polyps.
Bowel resection without reduction was entertained, encompassing the intussuscepted ileum as well as the right colon up to the hepatic flexure.
The specimen was inspected in the operating room. The resected ileum was roughly 30 cm in length and was harboring a grape-like set of polyps as lead point. In addition a 2 cm polyp was developing from the base of the appendix which was also intussuscepted into the right colon, congested and larger than the surgeons thumb. The margins of resection were free of polyps and the intraoperative gross exam showed no signs of malignancy.
The patient's postoperative course was uneventful. She recovered quickly and was discharged on the fifth postoperative day.
Pathology report confirmed the amartomatous nature of all polyps and the abscence of cancer or carcinoid.

Discussion

Unlike the pediatric population, intestinal intussusception is quite uncommon in adults. It has been estimated that 5% of intussusceptions occur in adults and it accounts for up to 5% of of alla cases of bowel obstruction of adults (5-11).
The etiology of the latter also differs, being well defined in more than 90% of patients and unknown in the remaining cases (1-5).
Finally, the treatment is different with 40 to 80% of pediatric cases treated by pneumatic or hydrostatic reduction and the diagnosis of intussusception in adults more often established in the operating room (2).
We will limit our discussion to the adult's intestinal intussusception.
Sex distribution among adults does not show any predilection. The median age of presentation is the sixth to seventh decade of life (1-5).
The lead point causing the intussusception can be found in both the small (64-81%) and the large bowel. Peristalsis and ingested food push the lesion caudad, dragging the adjacent normal bowel (intussusceptum) into the relaxed segment of distal intestine (intussuscipiens) (2).
Benign as well as malignant lesions have been reported as cause of adult intestinal intussusception.
Primary adenocarcinomas are responsible for almost all the large bowel intussusceptions whereas primary, malignant small bowel tumors rarely if ever present as intussusception (14).
Cases of cytomegalovirus colonic intussusception have been rarely reported.
Small bowel intussusceptions are more often caused by benign lesions (55%) and when malignant they are metastatic from sarcoma, melanoma, and lung cancer (1-6).
The clinical presentation may vary from the acute onset with signs and symptoms of intestinal obstruction to more subtle and chronic forms in which symptoms may even last 1 year, being represented only by abdominal pain (1-5).
The physical findings are usually those of bowel obstruction including a possible palpable mass (1-5).
The evaluation of these patients usually starts with plain radiographs of the abdomen, often showing air-fluid levels. Both contrast studies, small bowel series and barium enema, can be performed and can be very helpful not only in defining the nature of the bowel obstruction but also to reduce the intussusception (2, 3, 13).
The use of CT scan is being more frequently used in the diagnostic evaluation of these patients and appears very useful in making the diagnosis of intussusception.
Ultrasound is not a very helpful tool in making a diagnosis of intussusception.
Colonoscopy is now more frequently and successfully used (1).
If the diagnosis is made before surgery and the patient is stable without signs of bowel ischemia, hydrostatic reduction may be considered. Otherwise if the initial work-up is consistent with intestinal obstruction no further evaluation is needed and surgery must be undertaken (2).
Attention has to be paid to rehydration and prompt surgical treatment without any additional delay (1-2).
Whether to reduce or resect the intussuscepted bowel without reduction is a matter of controversy (1-2).
Resection without reduction is based on the rational that intraluminal seeding or venous embolization of a malignant etiology can be prevented. Anastomotic complications in the setting of an edematous bowel wall can also be avoided (4).
Unnecessary small bowel resection can expose the patient to the risk of a short gut syndrome especially when the underlying cause of intussusception is a Peutz-Jeghers syndrome (1).
In this case the intussusception may recur and multiple small bowel resection may eventually lead to a short gut. Reduction of an intussuscepted small bowel appears reasonable in light of the most frequent benign etiology. Rese-

ction without reduction seems the most appropriate treatment when a section of the large bowel is involved.
In our patient after the initial hemodynamic stabilization and diagnostic evaluation we decided to explore the patient. A long but unremarkable history of Peutz-Jeghers with the onset of a never experienced clinical picture of bowel obstruction was suggestive for a lost of balance between the patient and her disease. Procrastination of a laparotomy to pursue any other test seemed inappropriate. The proximity of the lead point to the ileocecal valve, the involvement of the cecum and the appendix, mandated a resection of a yet limited portion of small bowel. Since we could not rule out a malignancy or a carcinoid we elected to extend the resection to the right colon.

Conclusion

Because of its rarity adult intussusception is a challenging condition that requires preoperative clinical skill and considerate intraoperative judgment.
The diagnosis more often relays on clinical judgement and once the patient has been stabilized surgery is the only way to diagnose and treat these patients.
Whether to resect or reduce the intussuscepted bowel still remains controversial. Current evidences seem to support resection without reduction for colonic intussusception, given the higher incidence of neoplastic etiology. A small bowel resection can be performed if after an initial reduction a tumor is evident.

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Commento Commentary

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Quadri clinici rari come l'invaginazione intestinale nell'adulto sono sempre interessanti perché obbligano a rimettere in discussione tutto quello che l'esperienza e l'uso hanno consolidato riflettendo criticamente sulle indicazioni chirurgiche. Molto interessante in questo lavoro la accurata analisi dei rischi e delle indicazioni al trattamento resettivo, analisi che comprende sia i quadri "benigni" a partenza dall'intestino tenue che quelli prevalentemente dovuti ad adenocarcinomi a partenza dal grosso intestino (1-2). Gli AA mettono giustamente in guardia sia da un eccesso di conservatorismo, che potrebbe riportare in tempi brevi ad una recidiva del quadro ostruttivo, così come da un eccesso di "aggressività". La distribuzione infatti della malattia, in questo caso la poliposi di Peutz Jeghers, su tutta la superficie intestinale, potrebbe in tempi successivi condurre a recidive del quadro ostruttivo e riresezioni, con il rischio gravissimo di un sindrome da intestino corto, altrettanto se non più invalidante della malattia che si è voluto trattare.

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