ANNALI ITALIANI DI CHIRURGIA

Pubblicazione on-line
ISSN 2239-253X
Direttore Nicola Picardi

Ann. Ital. Chir.
e-publish 12 September 2012
www.annitalchir.com - pii: S2239253X12019081

Ileocecal-colonic intussusception caused by cecal adenocarcinoma

A case report

Luigi Verre, Rachele Rossi, Ilaria Gaggelli, Alessandro Piccolomini, Vlasta Podzemny, Andrea Tirone

U.O.C. Chirurgia 2 (Chair: A.F.Carli), AOU Senese, Siena, Italy

Ileocecal-colonic intussusception caused by cecal adenocarcinoma. A case report

INTRODUCTION: *Intussusception in adults is an infrequent cause of intestinal occlusion that is usually due to neoplasm lesions. The unspecific nature of the clinical presentation often delays diagnosis. It is most commonly emergency explorative laparotomy which clarifies the etiology of the occlusion. The authors report a case of intestinal occlusion caused by ileocecal-colonic invagination with a small cecal adenocarcinoma as lead point, in a 74-year-old woman.*
CASE REPORT: *A 74-year-old woman came to the Emergency Department, complaining of crampy pain in the mid- and lower abdomen. An abdominal ultrasound revealed a "pseudokidney sign" apparently involving the cecum. Because there were no clear signs of occlusion, the patient was dicharged on the same day. Three days later, upon admission, the patient complained of episodes of abdominal pain with intervals of moderate well-being, associated with nausea, vomiting and an inability to pass stool (but not gas) for 36 hours. On clinical examination her abdomen was distended and tender on palpation in all quadrants, especially in the right iliac fossa where a large mass could be felt. Standard abdominal x-Ray documented gaseous distension of some loops of the jejunum-ileum with some air-fluid level. The patient underwent an abdominal CT scan which showed advanced intussusception that appeared to be ileocolic and multiple enlarged lymphnodes were found in the invaginated mesentery at the base of which there appears to be a thickening of the intestinal wall that is probably neoplastic in nature. The patient underwent explorative laparotomy. Ileocecal-colonic intussusception caused by a cecal growth 5 cm in diameter was found on examination of the surgical specimen. Histology showed that the cause of the large swelling of the ascending colon was a vegetating ulcerated adenocarcinoma (medium grade differentiation: G2), measuring 6.5x 4.0 cm, arising from a tubulovillous adenoma infiltrating the submucosa.*
CONCLUSIONS: *Most cases of intussusception are caused by structural lesions, a large percentage of which are malignant, especially in the colon. In our patient the lead point was a small cecal polyp which, together with the last loop of the ileum and the ileocecal valve, was pulled into the ascending colon. Although most cases of intussusception in adults are diagnosed at the operating table, noninvasive diagnostic tools like ultrasonography and CT scanning are very useful. Treatment in adults is usually surgical and involves en bloc resection of the lesion. Manual reduction of the intussusception is not advisable because of the risk of dissemination if the lead point is malignant.*

KEY WORDS: Cecal adenocarcinoma, Itestinal resection, Intussusception in adults.

Introduction

Intussusception is rare in adults and is almost always associated with a polyp, tumor, or Meckel's diverticulum [1]

Nonspecific symptoms, similar to those associated with other causes of intestinal occlusion often make it difficult to diagnose intussusception. Computed tomography (CT) scanning and ultrasound examination provide important information about the location of the intestinal segment involved and sometimes about the mechanism (i.e.: volvulus, invagination). However neither investigation can identify the nature of the lesion and so, in most cases, the diagnosis is established by means of explorative laparotomy. Management of ileocolic or colocolonic intussusception always necessitates *en bloc* resection of the segment involved due to the high incidence

Pervenuto in Redazione Marzo 2012. Accettato per la pubblicazione Maggio 2012
Correspondence to: Andrea Tirone, Policlinico Le Scotte UOC Chirurgia 2, Viale Bracci 53100 Siena, Italy (e-mail:andrea.tirone@alice.it)

of malignant neoplasia and intestinal ischemia associated with this pathology [2,3]. The case of intussusception presented here, with a small cecal adenocarcinoma as lead point, serves to focus attention on the issue of diagnosis and treatment of cases of intestinal occlusion that are hard to interpret, and especially on the time required to make a diagnosis and the best treatment approach [4].

Case report

A 74 year old woman came to the Emergency Department, at the suggestion of her general practitioner, complaining of crampy pain in the mid- and lower abdomen that had begun one week prior to presentation. An abdominal x-ray showed "a slight distension of some loops of the ileum but no air-fluid levels, and air around the colon without signs of perforation".
Abdominal ultrasound revealed a "pseudokidney sign" apparently involving the cecum (Fig. 1).
Blood test results were normal except for mild leucocytosis (12,280 /mm3). Because there were no clear signs of occlusion, the patient was dicharged on the same day and advised to return in 3 days for an abdominal x-ray (after fasting) and investigative colonoscopy.
Three days later, upon admission, the patient complained of episodes of abdominal pain with intervals of moderate well-being, associated with nausea, vomiting (bile-stained material containing mucus) and an inability to pass stool (but not gas) for 36 hours. On clinical examination her abdomen was distended and tender on palpation in all quadrants, especially in the right iliac fossa where a large mass could be felt (Fig. 2).

Fig. 1: "Pseudokidney" sign apparently involving the cecum.

Fig. 2: The abdominal mass in the right iliac fossa.

As a temporary measure for intestinal decompression a nasogastric tube was positioned and 500 ml of bile-like fluid was aspirated. Blood tests revealed mild neutrophilic leucocytosis (16,000/mm^3), hyponatremia (128 mEq/l), and hypokalemia (3.3 mEq/l). Standard abdominal x-Ray documented "gaseous distension of some loops of the jejunum-ileum with some air-fluid levels, but without pneumoperitoneum or air in the rectal ampulla".
In the light of these findings the patient underwent an abdominal CT scan which showed "advanced intussusception that appeared to be ileocolic in a patient with dolichocecum without signs of vascular involvement. Multiple enlarged lymphnodes were found in the invaginated mesentery at the base of which there appears to be a thickening of the intestinal wall that is probably neoplastic in nature" (Figg. 3, 4)
After restoration of the fluid and electrolyte balance the patient underwent explorative laparotomy to identify the cause of the intestinal occlusion.
Ileocecal-colonic intussusception caused by a cecal growth 5 cm in diameter was found on examination of the surgical specimen. (Fig. 5)
Ileocecal resection with ileocolic laterolateral mechanical anastomosis was therefore required.
The patient's postoperative course was uneventful except for an episode of atrial flutter which resolved after pharmacological treatment. The nasogastric tube was removed on day 2 and the abdominal drain on day 6. The patient

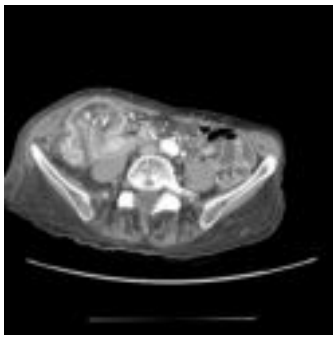

Fig. 3: Advanced ileocolic intussusception and medially displaced dolichocecum.

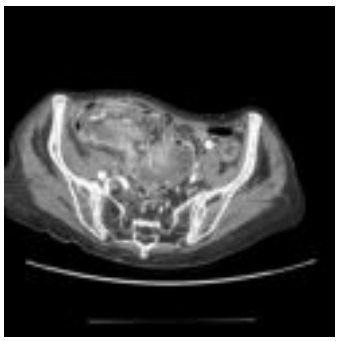

Fig. 4: Ileocolic intussusception with invaginated mesentery and thickening of the intestinal wall seemingly due to heteroplasia.

Fig. 5: Two views of the surgical specimen.

had her first postoperative bowel movement on day 4.
Histology showed that the cause of the large swelling of the ascending colon, resulting from ileocecal-colonic intussusception, was a vegetating ulcerated adenocarcinoma (medium grade differentiation: G2), measuring 6.5x 4.0 cm, arising from a tubulovillous adenoma infiltrating the submucosa.
The patient was discharged on day 8 and will be followed up as an outpatient according to our follow up protocol used for 700 patients operated on for colon cancer.

Discussion

Intussusception occurs when a segment of the intestine with its mesentery (the intussusceptum) invaginates (slides) into the lumen of an adjacent segment (the intussuscipiens) [5].
In contrast to intussusception in infants, most intussusception in adults is caused by well-defined lesions or structural changes ([1-13]). Neoplasia causes 2/3 of the cases of intussusception in adults and 60% of these lesions are malignant [14], especially those located in the colon. The remaining third are due to inflammatory diseases, appendicits, lymphoid hyperplasia, anastomoses, and suture lines.
Most causes of intussusception in the ileum are benign:Meckel's diverticulum, adhesions, and celiac disease [2,3,10-12,15]. In past few decades cases of ileal intussusception associated with autoimmune deficiency syndrome (AIDS) have also been reported [16-20] with the lead point due to lymphoma, Kaposi's syndrome, reactive lymphoid hyperplasia, atypical mycobacterial infection, cytomegalovirus (CMV)-induced colitis, and Campylobacter-related enteritis. Intussusception should always be included in the differential diagnosis if AIDS patients present with episodes of abdominal pain and symptoms of intestinal occlusion.
In tropical regions a lot of cases of intussusception are due to endemic enteric infections, which are associated with diarrhea, disorders of peristalsis, and a high risk of invagination [21-25].
In adults intussusception affects men and women equally and most commonly occurs in individuals around 50 years of age.
Although in children there are usually acute symptoms and characteristic signs, for instance rectal discharge (currant jelly stools), in adults there are mostly nonspecific symptoms which can be subacute or chronic. The most common are crampy abdominal pain, nausea and vomiting. Less frequently there is diarrhea or constipation. Rectal bleeding and a palpable abdominal mass are found only in a minority of cases.
Some patients are asymptomatic, for instance those with celiac disease whose intestinal muscles are too weak too cause abdominal cramps [2,8,10-12,14, 23,26-28].
Making a clinical diagnosis of intussusception in an adult is always a challenge because the condition is uncommon and the symptoms usually nonspecific. Some studies report that a correct preoperative diagnosis was made in 32-50% of cases and that it was easier to diagnose patients with malignant colonic lesions than those with benign intestinal lesions (67% vs. 22%) [2,3,15].
An abdominal x-ray is often the first diagnostic investigation performed in a patient who presents with symptoms of intestinal occlusion. It is useful for evaluating and monitoring the degree of occlusion and identifying any signs of pneumoperitoneum.
Abdominal ultrasound examination is useful in diagnosing intussusception in adults, especially when a palpable abdominal mass is present and accessible to examination, as in our patient [29-33]. In particular, when the probe is held transverse to the intussusception, the typical image is that of a mass (target) with a hypoechogenic border (due to the edema in the wall of the intussuscipiens) and a central echogenic area (the intussusceptum and the fat jn the invaginated mesentery). When an oblique section of the intussusception is visualized, it is called a "pseudokidney sign": the edematous intestinal wall mimics the hypoechogenic renal cortex and the hyperechogenic intussusceptum mimics the renal sinus [29].
However, although they are suggestive of intussusception, these lesions are not pathognomic and can also be observed in other cases of intestinal wall edema, for instance edema caused by enterocolitis or volvulus
In recent years CT scanning has become more and more widely used in diagnosing patients with nonspecific abdominal pain because it identifies pathognomic signs of intussusceptions [27,34-37]: the area of intussusception appears like a lesion due to thickening of the intestinal

wall (intussuscipiens and intussusceptum) with a central zone of adipose tissue showing vascular enhancement (mesenterial fat).
This gives the area a target-like appearance.The layering can be due to trapping of fluid between the intussusceptum and the intussuscipiens or to edema in the wall of the intussuscipiens [5,38]. Although it is possible to make a fairly certain diagnosis based on a CT scan, it is difficult to determine the underlying etiology [18,34] since it is not easy to distinguish between the neoplastic mass and the intussusception itself. The presence of pathological lymph nodes or metastases on a CT scan can indirectly reveal the presence of a malignant tumor. However these details cannot be considered definite indications that the lead point is malignant. In the literature there are reports that decribe intussusception not due to cancer in patients with known malignancy [34,39].
Another diagnostic investigation that is useful for visualizing intussusception is magnetic resonance imaging (MRI) [27,40].
Even though in children.non -surgical reduction with barium or air enemas seems to be effective for intussusception, there is still debate regarding the correct treatment for adults. It seems to be generally accepted that in adults surgery is obligatory due to the high incidence of malignant lead points and the severe complications of intestinal occlusion and intestinal ischemia [4,41] and *en bloc* resection of the lesion is the treatment of choice since manipulation of the lesion during reduction increases the risk of intraluminal dissemination, venous embolism in the tumor itself, or perforation when there is intestinal ischemia [12,15].
Surgical intervention in cases of intussusception with a benign etiology such as celiac disease and in case of transient intussusception (a short segment, nonocclusive) found incidentally on CT scans in relatively asymptomatic patients, is absolutely contraindicated [28].

Conclusions

Intussusception is relatively rare in adults and differs substantially from pediatric intussusception. Most cases of intussusception are caused by structural lesions, a large percentage of which are malignant, especially in the colon. In our patient the lead point was a small cecal polyp which, together with the last loop of the ileum and the ileocecal valve, was pulled into the ascending colon.
Although most cases of intussusception in adults are diagnosed at the operating table, noninvasive diagnostic tools like ultrasonography and CT scanning are very useful. In our case intussusception was already suspected after ultrasound examination.
Treatment in adults is usually surgical and involves *en bloc* resection of the lesion. Manual reduction of the intussusception is not advisable because of the risk of dissemination if the lead point is malignant. The intussusception in our patient was therefore resected and then opened, revealing a macroscopic lesion on the wall of the colon
However when there is a benign cause underlying invagination surgical management is completely inappropriate.

Riassunto

L'intussuscezione intestinale nell'adulto rappresenta una condizione occlusiva non frequente e per lo più sostenuta da lesioni patologiche. La non specificità del quadro clinico spesso ritarda la diagnosi ed è solitamente la laparotomia esplorativa d'urgenza a chiarire l'eziologia del quadro occlusivo sebbene esistano esami diagnostico-strumentali,come la TC, che offrono utilissime informazioni preoperatorie. Gli Autori riportano un caso di occlusione intestinale eseguito in regime d'urgenza e sostenuto da una invaginazione ileo-ceco-colica su un piccolo adenocarcinoma del cieco, in una donna di anni 74, presentatasi al Pronto Soccorso con un quadro di dolore addominale ed una massa palpabile in fossa iliaca destra. Nel caso descritto l'invaginazione intestinale, ingenerata da un piccolo adenocarcinoma del cieco, ci ha permesso di analizzare la complessa problematica dell'iter diagnostico e terapeutico dei casi di occlusione intestinale di difficile interpretazione, focalizzando l'attenzione sulla corretta tempistica della diagnosi e sull'adeguatezza dei provvedimenti terapeutici adottati.

References

1. Agha FP: *Intussusception in adults*. AJR Am J Roentgenol, 1986; 146:527-31.

2. Azar T, Berger DL.: *Adult Intususception*. Ann Surg, 1997; 226: 134-48.

3. Begos D, Sandor A, Modlin I: *The diagnosis and management of adult intususception.* Am J Surg, 1997; 173:88-94.

4. Zubaidi A, Al- Saif F, Silverman R.: *Adult intususception: A retrospective review.* Dis Colon Rectum, 2006; 49:1546-551.

5. Huang BY, Warshauer DM: *Adult intussusception: Diagnosis and clinical relevance*. Radiol Clin N Am, 2003; 41:1137-151.

6. Brayton D, Norris WJ: *Intussusception in adult*. Am J Surg, 1954; 88:32-43.

7. Courtney SP, Ibrahi *[see comments]*. Postgrad Med J, 1992; 68:449-52.

8. Dean D, Ellis F, Sauer W: *Intussusception in adults*. Arch Surg, 1956; 73:6-11.

9. Donhauser JL, Kelly EC: *Intussusception in the adult*. Am J Surg, 1950; 79:673-77.

10. Eisen L, Cunningham J, Aufses A: *Intussusception in adults: Institutional review*. J Am Coll Surg, 1999; 188:390-95.

11. Hamaloglu E, Yavuz B: *Intussusception in adults*. Panminerva Med, 1990; 32:118-21.

12. Nagorney DM, Sarr MG, McIlrath DC: *Surgical management of intussusception in the adult.* Ann Surg, 1981; 193:230-36.

13. Nicolas JC, Ingrand D, Fortier B, Bricout F: *A oneyear virological survey of acute intussusception in childhood.* J Med Virol, 1982; 9:267-71.

14. Felix EL, Cohen MH, Bernstein AD, Schwartz JH.: *Intussusception; Case report of recurrent intussusceptions and review of the literature*. Am J Surg, 1976; 131:758-61.

15. Reijnen HA, Joosten HJ, de Boer HH: *Diagnosis and treatment of adult intussusceptions.* Am J Surg, 1989; 158:25-28.

16. Cappell MS, Hassan T, Rosenthal S, Mascarenhas M: *Gastrointestinal obstruction due to Mycobacterium avium intracellulare associated with the acquired immunodeficiency syndrome*. Am J Gastroenterol, 1992; 87:1823-827.

17. Danin JC, McCarty M, Coker R: *Case report: Lymphoma causing small bowel intussusception in a patient with the acquired immune deficiency syndrome.* Clin Radiol, 1992; 46:350-51.

18. Meyerson S, Desai TK, Polidori G, Raval MF, Ehrinpreis MN: *A case of intussusception and lymphoid hyperplasia in a patient with AIDS" [see comments].* Am J Gastroenterol, 1993; 88:303-06.

19. Visvanathan R, Nichols TT, Reznek RH: *Acquired immune deficiency syndrome-related intussusception in adults*. Br J Surg, 1997; 84:153-40.

20. Wood BJ, Kumar PN, Cooper C, Silverman PM, Zeman RK: *AIDS-associated intussusception in young adults*. J Clin Gastroenterol, 1995; 21:158-62.

21. Ademiluyi SA: *Intussusception in Nigerian adults*. J Natl Med Assoc, 1987; 79:873-76.

22. Altri SC, Singh AK, Naithani YP: *Adult intussusception in Allahabad*. J Indian Med Assoc, 1983; 81:155-85.

23. Coleman MJ, Hugh TB, May RE, Jensen MJ: *Intussusception in the adult*. Aust N Z J Surg, 1981; 51:179-80.

24. Hadley GP, Simpson RL: *Adult intussusception in the tropics*. Br J Surg, 1983; 70:281.

25. Otu AA: *Tropical surgical abdominal emergencies: Acute intestinal obstruction*. Afr J Med Med Sci, 1991; 20:83-88.

26. Catalano O: *Transient small bowel intussusception: CT findings in adults.* Br J Radiol, 1997; 70:805-08.

27. Warshauer DM, Lee JK.: *Adult intussusception detected at CT or MR imaging: Clinical-imaging correlation.* Radiologym, 1999; 212:853-60.

28. Weilbaecher D, Bolin JA, Hearn D, Ogden WD: *Intussusception in adults: Review of 160 cases*. Am J Surg, 1971; 121:531-35.

29. Boyle MJ, Arkell LJ, Williams JT: *Ultrasonic diagnosi on intussusceptions (letter: see comments).* Am J Gastroentero, 1993; 88:617-18, 2000; 25:4-57.

30. Cerro P, Macrini L, De Angelis O: *Somographic diagnosis of intussusception in adults.* Eur Radiol, 2000; 10:811-13.

31. Sato M, Ishida H, Konno K, et al.: *Long-standing painless intussusception in adults*. Eur Radiol, 2000; 10:811-13.

32. Sofia S, Casali A, Bolondi L: *Sonographic diagnosis of adult intussusceptions.* Abdom Imaging, 2001; 26:483-86.

33. Weissberg DL, Scheible W, Leopold GR: *Ultrasonographic appearance of adult intussusceptions*. Radiology, 1977; 124:791-92.

34. Gayer G, Apter S, Hofmann C, et al.: *Intussusception in adults: CT diagnosis.* Clin Radiol, 1998; 53:53-57.

35. Gayer G, Zissin R, Apter S, Papa M, Hertz M: *Pictorial review: Adult intussusception: A CT diagnosis*. Br J Radiol, 2002; 75:185-90.

36 .Iko BO, Teal JS, Siram SM, Chinwuba CE, Roux VJ, Scott VF: *Computed tomography of adult colonic intussusception: clinical and experimental studies*. AJR Am J Roentgenol, 1984; 143:769-72.

37. Merine D, Fishman EK, Jones B, Siegelman SS: *Enteroenteric intussusception: CT findings in nine patients*. AJR Am J Roentgenol, 1987; 148:1129-132.

38. Karamercan A, Kurukahvecioglu O, Yilmaz TU: *Adult ileal intussusceptions: An unusual emergency condition*. Adv Thar, 2006; 23(1):163-68.

39. Lorigan JG, DuBrow RA: *The computed tomographic appearances and clinical significance of intussusceptions in adults with malignant neoplasms*. Br J Radiol, 1990; 63:257-62.

40. Marcos HB, Semelka RC, Worawattanakul S: *Adult intussusception: demonstration by current MR techniques*. Magn Reson Imaging, 1997; 15:1095-098.

41. Wang N, Cui XY, Liu Y, Long J, Xu HY, Guo RX, Guo KJ: *Adult intussusception: A retrospective review of 41 cases*. World J Gastroenterol, 2009, 15:3303-308.