

악성 림프종에 의한 성인에서의 회맹부 장중첩증

남상근¹, 강정현¹, 박효진², 이강영¹, 손승국¹

연세대학교 의과대학 ¹외과학교실, ²내과학교실

Adult ileocecal intussusception caused by malignant lymphoma

Sanggeun Nam¹, Jeonghyun Kang¹, Hyojin Park², Kang Young Lee¹, Seung-Kook Sohn¹

Departments of ¹Surgery and ²Internal Medicine, Yonsei University College of Medicine, Seoul, Korea

Adult intussusception is rare but can prove to be challenging surgical situation. The etiology of adult intussusception is different from that of children, and because of the possibility of malignancy, surgical treatment is highly recommended. We present a case report of malignant lymphoma that caused ileocolic and colocolic intussusception in an adult. At first event of intussusception, the patient refused to undergo additional treatments due to improved symptoms. Three months later, the patient was transferred to our hospital due to recurrent and unresolved abdominal pain. The patient underwent a right hemicolectomy and was diagnosed with malignant B-cell lymphoma that involved the ileocecal valve. Physicians should consider malignancy as a potential cause of intussusception in adult, and should quickly provide adequate surgical intervention.

Keywords: Intussusception, Ileocecal valve, Lymphoma, Adult

INTRODUCTION

Intussusception occurs when one segment of the bowel and associated mesentery invaginate into an adjacent segment [1]. Intussusception is primarily a childhood disease. The incidence of intussusception is 1.5 to 4 cases per 1,000 live births, and it often occurs around 1 year of age [2]. Adult intussusception is a rare clinical entity that accounts for 5% of all intussusceptions [3]. Vague and non-specific symptoms and signs often make a preoperative diagnosis difficult. It has been reported that 63% of adult intussusceptions are related to tumors [4]. Because of the high risk of malignancy, surgical intervention is recommended in cases of adult intussusception. We report a case of malignant lymphoma that caused intussusception in the ileocecal region.

Received: Apr 23, 2014 **Revised:** May 26, 2014 **Accepted:** Jun 20, 2014

Correspondence to: Jeonghyun Kang

Department of Surgery, Gangnam Severance Hospital, Yonsei University College of Medicine, 211 Eonju-ro, Gangnam-gu, Seoul 135-720, Korea

Tel: +82-2-2019-3369, **Fax:** +82-2-3462-5994

E-mail: ravic@naver.com

Copyright © Korean Society of Surgical Oncology

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

CASE REPORT

A 51-year-old female visited our hospital and reported three-day history of recurrent abdominal pain. She had no specific past medical or family history. One year earlier, she underwent a colonoscopy during a routine health checkup, which showed no abnormal findings. Three months earlier, she visited a local hospital because of right upper quadrant abdominal pain with a palpable mass. Although it was recommended that she receive additional treatment due to the suspicion of an intussusception, she refused to undergo additional procedures because the symptoms resolved spontaneously.

On physical examination, a palpable mass was noted in the right upper quadrant of the abdomen. Laboratory studies upon admission yielded normal chemistry results, but low hemoglobin levels (6.8 g/dL). The levels of carcinoembryonic antigen and carbohydrate antigen 19-9 were within normal limits. An abdominopelvic computed tomography (CT) scan showed a circumferential mural thickening and dilatation of the distal ileum. Mural thickening and enhancement of the dilated appendix (9.7 mm) in the invaginated large bowel lumen was noted (Fig. 1). Multiple enlarged mesenteric lymph nodes were also detected.

According to the CT images, the diagnosis was colocolic and ileocolic intussusception. Although there was no definite mass-like lesion in this

CT examination, there was a possibility of hidden malignancy at the ileocecal valve as the leading point of intussusception. We scheduled an elective exploratory-laparotomy, which indicated that the small intestine was invaginated through the ileocecal valve and that the mesenteric lymph nodes were enlarged (Fig. 2). Frozen section results of an enlarged lymph node revealed that it was a malignant lymphoma. A right hemicolectomy with end-to-side ileotransverse colostomy was performed.

After surgery, the patient recovered uneventfully. The final pathologic diagnosis indicated a diffuse large B cell-type malignant lymphoma (Fig. 3). Immunohistochemical staining revealed atypical lymphoid cells that were positive for CD20. Analysis indicated that malignant lymph nodes were involved in 5 out of 14 regional lymph nodes. She was transferred to the hematology-oncology department and underwent 6 cycles of rituximab-cyclophosphamide, doxorubicin, vincristine, and prednisone chemotherapy.

DISCUSSION

Intussusception occurs when a proximal portion of the bowel invagi-

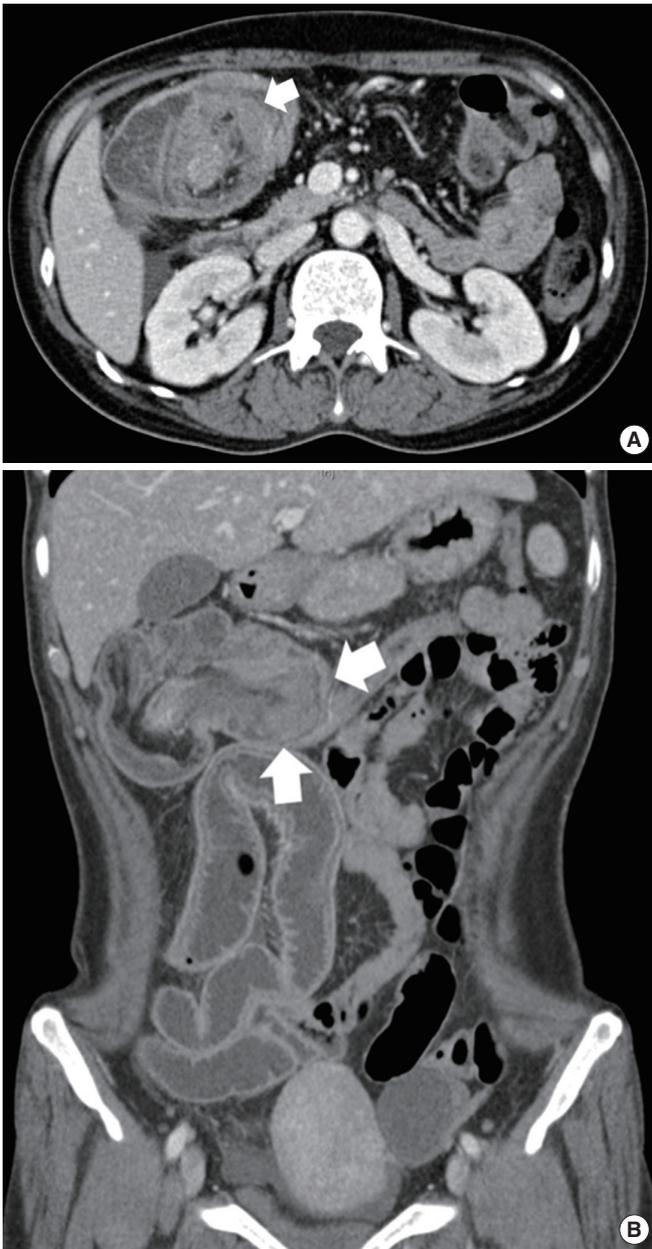


Fig. 1. (A) Abdominal computed tomography showing a target lesion with circumferential mural thickening and dilatation of the distal ileum, suggesting ileocolic intussusception (arrow). (B) Invagination of the terminal ileum, segmental distal ileum, and cecum into the lumen of the proximal transverse colon (arrow).

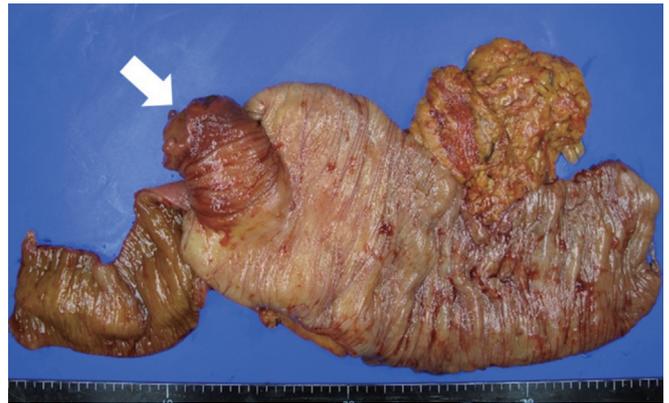


Fig. 2. Resected specimen showing intussusception. The small intestine had invaginated through the ileocecal valve (arrow, invaginated small bowel located in the cecum).

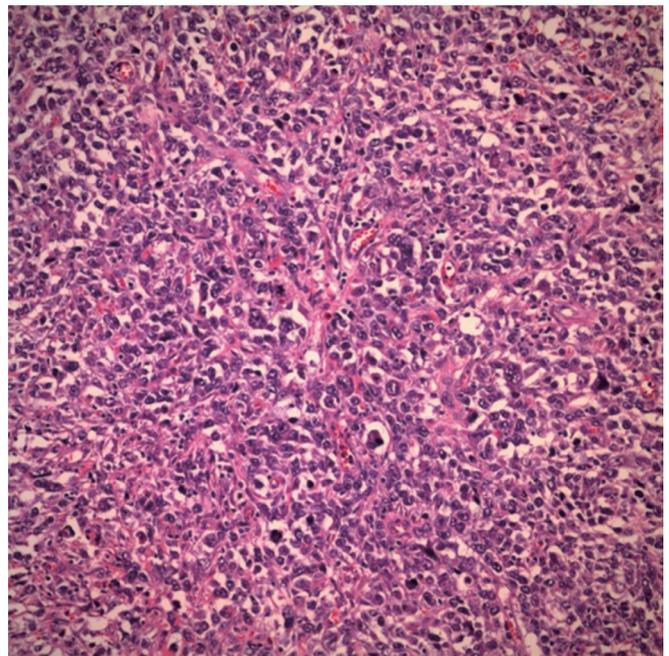


Fig. 3. Photomicrograph showing B-cell lymphoma cells in the intestinal mucosa (H&E, x200).

nates into the more distal bowel [1]. Although intussusception is a major cause of intestinal obstruction in children, it is rare in adults. Adult intussusception represents only 5% of all intussusceptions [4], and common clinical presentations in adult patients with intussusception include abdominal pain, nausea, vomiting, and abdominal distension [3,5]. However, the preoperative diagnosis of adult intussusception is very difficult because the typical symptoms such as pain, a palpable mass, and bloody stool, are present only in 10% of the patients [6]. In a review of 1,214 reported cases of intussusception in adults, Felix et al. [4] estimated that 55% (668/1,214) involved the small intestine, whereas 45% (546/1,214) involved the colon. Of the colon intussusceptions, 48% resulted from malignant tumors and 21% from benign lesions [4]. Begos et al. [7] reported that intussusception of the small intestine in adults is due to benign (63%), idiopathic (23%), and malignant (14%) lesions.

It is well known that the gastrointestinal tract is the most common site for extranodal non-Hodgkin lymphoma, accounting for about 20% to 50% of these lymphomas [3]. However, large bowel involvement is rare, and occurs in only 10% of gastrointestinal presentations of malignant lymphoma. The cecum is the lesion site in 85% of cases of large bowel malignant lymphoma [8,9]. Akbulut [3] reported that the ileocolic type was the most common (66.6%, 24/36), followed by the enteric type (27.7%, 10/36) among 36 cases of intussusceptions due to lymphoma that were reported in the literature from 2000 to 2011. In our patient, the final pathologic results revealed that ileocolic lymphoma caused an ileocolic intussusception.

In adult intussusception that involves the colon, most cases originated from an anatomical leading point. Therefore, surgical treatment should not be delayed for adult intussusception. Currently, there is some debate regarding when to perform the reduction procedure. If a reduction could be performed successfully, it could be possible to perform a more limited resection. However, reduction should not be recommended in cases of inflammation or ischemic change of the bowel [10]. When the possibility of malignancy cannot be excluded, tumor spillage during the reduction process could be fatal for the patient. In addition, spontaneous reduction could not guarantee the absence of a pathologic cause.

In this case, the patient had previously visited the emergency department of another hospital for diffuse abdominal pain 3 months earlier and was diagnosed with an intussusception. Because the discomfort was not severe and it disappeared spontaneously, she refused to undergo further treatment at that stage. Although the intussusception could spontaneously disappear after the first episode of the event, the patient could have been treated earlier if she had been strongly informed of the possibility of a hidden malignancy. Interestingly, the patient underwent colonoscopy for a health check-up one year before the visit to our hospital. According to the patient's statements, the colonoscopy did not reveal

any abnormalities. Therefore, it could be possible to identify ileocolic lymphoma at an earlier stage with a thorough evaluation of the terminal ileum, although it is uncertain whether the performance of ileocecal valve intubation during the colonoscopy was performed. Thus, this case emphasizes that physicians should be more cautious of the possibility of malignancy in adult intussusception.

In conclusion, we report a case of malignant lymphoma in the ileocecal region that caused colocolic intussusception in an adult. Physicians should be more suspicious of malignancy for an intussusception of unknown cause in adult patient. Finally, immediate surgical intervention is highly recommended for these patients.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

- Huang BY, Warshauer DM. Adult intussusception: diagnosis and clinical relevance. *Radiol Clin North Am* 2003;41:1137-51.
- Fischer TK, Bihmann K, Perch M, Koch A, Wohlfahrt J, Kare M, et al. Intussusception in early childhood: a cohort study of 1.7 million children. *Pediatrics* 2004;114:782-5.
- Akbulut S. Unusual cause of adult intussusception: diffuse large B-cell non-Hodgkin's lymphoma: a case report and review. *Eur Rev Med Pharmacol Sci* 2012;16:1938-46.
- Felix EL, Cohen MH, Bernstein AD, Schwartz JH. Adult intussusception; case report of recurrent intussusception and review of the literature. *Am J Surg* 1976;131:758-61.
- Loukas M, Pellerin M, Kimball Z, de la Garza-Jordan J, Tubbs RS, Jordan R. Intussusception: an anatomical perspective with review of the literature. *Clin Anat* 2011;24:552-61.
- Wang N, Cui XY, Liu Y, Long J, Xu YH, Guo RX, et al. Adult intussusception: a retrospective review of 41 cases. *World J Gastroenterol* 2009;15:3303-8.
- Begos DG, Sandor A, Modlin IM. The diagnosis and management of adult intussusception. *Am J Surg* 1997;173:88-94.
- Salemis NS, Tsiambas E, Liatsos C, Karameris A, Tsohataridis E. Small bowel intussusception due to a primary non-Hodgkin's lymphoma. An unusual presentation and clinical course. *J Gastrointest Cancer* 2010;41:233-7.
- Ghimire P, Wu GY, Zhu L. Primary gastrointestinal lymphoma. *World J Gastroenterol* 2011;17:697-707.
- Tan KY, Tan SM, Tan AG, Chen CY, Chng HC, Hoe MN. Adult intussusception: experience in Singapore. *ANZ J Surg* 2003;73:1044-7.